Improving the mental health of children and young people with long term conditions: Linked evidence syntheses

Darren A Moore[1], Michael Nunns[1†], Liz Shaw[1†], Morwenna Rogers[2], Erin Walker[3], Tamsin Ford[4], Ruth Garside[5], Obi Ukoumunne[2], Penny Titman[3], Roz Shafran[6], Isobel Heyman[3], Rob Anderson[1], Chris Dickens[2], Russell Viner[6], Sophie Bennett[6], Stuart Logan[2], Fiona Lockhart[7], Jo Thompson Coon[2*]

[1]Evidence Synthesis & Modelling for Health Improvement, University of Exeter Medical School, Exeter, UK

[2]National Institute for Health Research (NIHR) Collaboration for Leadership in Applied Health Research and Care South West Peninsula (PenCLAHRC), University of Exeter Medical School, Exeter, UK

[3]Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK

[4]Child Mental Health Group, University of Exeter Medical School, Exeter, UK

[5]The European Centre for Environment and Human Health (ECEHH), University of Exeter Medical School, Truro, UK

[6]University College London Institute of Child Health, London, UK

[7]Biomedical Research Centre Patient & Public Involvement Group, University College London Hospitals, London, UK

[*]Corresponding author: J.Thompson-Coon@exeter.ac.uk

[†]Joint second author

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Abstract

Background

Although mental health difficulties can severely complicate the lives of children and young people (CYP) with long-term physical conditions (LTCs), there is a lack of evidence about the effectiveness of interventions to treat them.

Objectives

To evaluate the effectiveness and cost-effectiveness of interventions aiming to improve the mental health of CYP with LTCs (Review 1), and explore the factors that may enhance or limit their delivery (Review 2).

Data Sources

For Review 1, 13 electronic databases were searched, including MEDLINE, Embase, PsycINFO, CENTRAL, CINAHL and Science Citation Index. For Review 2, MEDLINE, PsycINFO and CINAHL were searched. Supplementary searches, author contact and grey literature searches were also conducted.

Review Methods

The first systematic review sought randomised controlled trials (RCTs) and economic evaluations of interventions to improve elevated symptoms of mental ill health in CYP with LTCs. Effect sizes for each outcome were calculated post-intervention (Cohen's *d*). Where appropriate, random-effects meta-analysis produced pooled effect sizes (*d*).

Review 2 located primary qualitative studies exploring experiences of CYP with LTCs, their families and/or practitioners, regarding interventions aiming to improve the mental health and wellbeing of CYP with LTCs. Synthesis followed the principles of meta-ethnography.

An overarching synthesis integrated the findings from Review 1 and Review 2 using a deductive approach. End-user involvement, including topic experts, CYP with LTCs and their parents, was a feature throughout the project.

Results

Review 1 synthesised 25 RCTs evaluating 11 types of intervention, sampling 12 different LTCs. Tentative evidence from seven studies suggests cognitive-behavioural therapy interventions could improve the mental health of CYP with certain LTCs. Intervention-LTC dyads were diverse, with few opportunities to meta-analyse. No economic evaluations were located.

Review 2 synthesised 57 studies, evaluating 21 types of intervention. Cancer, HIV positive and mixed-LTC samples were most common. Interventions often aimed to improve broader mental health and wellbeing, rather than symptoms of mental health disorder. The meta-ethnography identified five main constructs, described in an explanatory line-of-argument model of the experience of interventions.

Nine overarching synthesis categories emerged from the integrated evidence, raising implications for future research.

Limitations

Review 1 conclusions were limited by the lack of evidence about intervention effectiveness. No relevant economic evaluations were located. There were no UK studies included in Review 1, limiting the applicability of findings. The mental health status of participants in Review 2 was usually unknown, limiting comparability with Review 1. The different evidence identified by the two systematic reviews challenged the overarching synthesis.

Conclusions

There is a relatively small amount of comparable evidence for the effectiveness of interventions for the mental health of CYP with LTCs. Qualitative evidence provided insight into the experiences that intervention deliverers and recipients valued. Future research should evaluate potentially effective intervention components in high quality RCTs integrating process evaluations. End-user involvement enriched the project.

Study registration

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List of abbreviations

Abbreviation	Definition
AAABIQ	Acceptance and Action for ABI Questionnaire
ABI	Acquired brain injury
ACT	Acceptance and Commitment Therapy
ACT	Asthma Control Test
ADHD	Attention-deficit/hyperactivity disorder
ADIS-IV-C/P	Anxiety Disorders Interview Schedule for DSM-IV: Parent and Child Versions
AIDS	Acquired immune deficiency syndrome
BAI	Beck Anxiety Inventory
BASC-2	Behaviour Assessment System for Children - Second Edition
BDI	Beck Depression Inventory
BDI-II	Beck Depression Inventory 2
BIFI-A	Brain Injury Family intervention for Adolescents
BOC	Best of Coping
BSI-18	Brief Symptom Inventory-18 item version
C/ASWS	Child/Adolescent Sleep-Wake Scale
CAMHS	Child and Adolescent Mental Health Services
CASI	Children's Anxiety Sensitivity Index
CBCL	Achenbach Child Behaviour Checklist
CBT	Cognitive behavioural therapy
CDI	Child Depression Inventory
CDI-CP	Children’s Depression Inventory-Child and Parent version
CDI-P	Child Depression Inventory – Parent Report

CDI-S	Child Depression Inventory-Short form
CDRS-R	Children's Depression Rating Scale–Revised
CDS-A	Child Depression Scale Abbreviated
CES-D	Centre for Epidemiological Studies Depression scale
CF	Cystic fibrosis
CFD	Cranio-facial differences
CFS	Chronic fatigue syndrome
CG	Clinical Guidelines X
CGAS	Children's Global Assessment Scale
CHD	Congenital heart disease
ChIPS	Chronic Illness Peer Support
CI	Confidence Interval
CODI	Coping with a Disease Questionnaire
CP-QOL	Cerebral Palsy Quality of Life
CRD	Centre for Reviews and Dissemination
CSI	Children's Somatization Inventory
CT	Cannot Tell
CYP	Children and Young People
CYPAG	Children and Young People Advisory Group
CYPMHC	Children and Young People's Mental Health Coalition
DASS	Depression, Anxiety, Stress Scale
DMAP	Developmental Martial Arts Program
DoH	Department of Health
DQOL	Diabetes Quality of Life for Youth scale
DSM-5	Diagnostic and Statistical Manual of Mental Disorders – Fifth Edition

DSQ	Diabetes Stress Questionnaire
DSRS-C	Depression Self-Report Scale-Children
ECBI	Eyberg Child Behaviour Inventory
EIT	Emotional Intelligence Training
FACE-TC	Family-Centred Advanced Care Planning for Teens with Cancer
FAD	Family Assessment Device
FAP	Functional Abdominal Pain
FDI	Functional Disability Inventory
FEV1	Forced Expiratory Volume over one second
GHb	Total glycosylated haemoglobin
GHQ	General Health Questionnaire
GOSH	Great Ormond Street Hospital for Children NHS Foundation Trust
HADS	Hospital Anxiety and Depression Scale
HAMD	Hamilton Depression Scale
HbA1C	Percentage of haemoglobin that has become glycated
HHI	Herth Hope Index
HIV	Human Immunodeficiency Virus
HRQOL	Health Related Quality Of Life
HTA	Health Technology Assessment
IBD	Inflammatory bowel disease
IBD-SAS	Inflammatory Bowel Disease-Specific Anxiety Scale
ICD-10	International Statistical Classification of Diseases and Related Health Problems – Tenth Edition
IQ	Intelligence Quotient
JAP	Peer-reviewed journal article

JFM	Juvenile Fibromyalgia
JIA	Juvenile Idiopathic Arthritis
Kiddie-SADS-L	Kiddie Schedule for Affective Disorders and Schizophrenia–Lifetime Version
K-SADS-PL	Kiddie-Schedule for Affective Disorders and Schizophrenia-Present Version
LOT-R	The Life Orientation Test-Revised
LTC	Long-term physical condition
MBSR	Mindfulness-Based Stress Reduction
MH	Mental Health
MMP	Movie Making Programme
MTCD	Music Therapy Compact Disc
mYPAS	Modified Yale Preoperative Anxiety Scale
NDST	Non-Directive Supportive Therapy
NG	NICE Guidelines
NHIS	National Health Interview Survey
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
NR	Not Reported
OECD	Organisation for Economic Co-operation and Development
PAIRS	Pain and Impairment Relationship Scale
PANAS-C	Positive and Negative Affects Schedule-Child version
PASCET-PI	Primary and Secondary Control Enhancement Therapy – Physical Illness
PCDAI	Paediatric Crohn's Disease Activity Index
PCQ	The Pain Coping Questionnaire

PCSC	Perceived Control Scale for Children
PEDI	Paediatric Evaluation of Disability Inventory
PedsQL	Paediatric Quality of Life Inventory
PFSC	Persistent Functional Somatic Complaints
PMR	Progressive Muscle Relaxation
PPC	Parenting Problem Checklist
PPQ	Varni-Thompson Pediatric Pain Questionnaire
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PS	Parenting Scale
PSOC-SF	Parenting Sense of Competency Scale-Short Form
PSRM	Personal and Social Responsibility Model
PTC	Parenting Tasks Checklist
PTFQ	Parent Thoughts and Feelings Questionnaire
PUCAI	Paediatric Ulcerative Colitis Activity Index
QoL	Quality of Life
QOLIE31	Quality of Life in Epilepsy Inventory
RCT	Randomised Controlled Trial
RFFD	Risk Factors For Depression
RQI	Relationship Quality Index
SB	Spina bifida
SCARED	Screen for Child Anxiety Related Emotion Disorder
SCD	Sickle cell disease
SD	Standard Deviation
SDQ	Strengths and Difficulties Questionnaire
SED	Self-Efficacy for Diabetes

SF-36	Short Form 36 Health Scale
SMD	Standardised Mean Difference
SMT	Stress Management Training
SSTP	Stepping Stones Triple P
STAI	Spielberger State-Trait Anxiety Inventory
STAI-S	Spielberger State-Trait Anxiety Inventory-State subscale
STAI-T	Spielberger State-Trait Anxiety Inventory-Trait subscale
T1DM	Type 1 diabetes mellitus
TAPS	Treatment of Anxiety and Physical Symptoms
TAPS+IBD	Treatment of Anxiety and Physical Symptoms + Inflammatory Bowel Disease
TAU	Treatment As Usual
THITWGC	The Hole in the Wall Gang Camp
TMV	Therapeutic Music Video
TSK	The Tampa Scale of Kinesiophobia
TTG	Terrific Tuesday Group
VAS	Visual Analogue Scale
VR	Virtual Reality
WOC	Ways of Coping Checklist
YAP	Young Adult Programme
YMCA	Young Men's Christian Association
YPMHAG	Young Persons Mental Health Advisory Group

Plain English Summary

Many children and young people with a long term physical health condition also experience feelings of depression, anxiety and other mental health issues and this impacts on their day to day life, their family and others around them. This review investigates whether interventions (treatments, strategies and resources) can help children and young people with their mental health. We also reviewed studies that explored the attitudes and experiences of those involved in receiving or delivering similar interventions.

We found 25 studies that evaluated 11 different types of interventions from cognitive behavioural therapy (7 studies) to music therapy (1 study). There were some positive effects for the strategies tested on both mental health and other outcomes, but often small studies meant exact effects were uncertain. Many of the studies were not very well designed or carried out and differences between research designs meant it was hard to compare different studies.

We found 57 studies that explored experiences of interventions. Analysis suggested it was important that strategies involve building good relationships and are delivered in what feels like a safe space. Participants in studies tended to like interventions that provided social support and helped them feel better about living with a long term physical condition. Successful interventions were viewed to be accessible and engaging. These studies were often conducted well, but they focused on different interventions than the review evaluating how well interventions work.

There are many gaps where interventions have not been studied for certain long term physical conditions. Important outcomes like school attendance and looking after yourself were not measured. More carefully designed UK research consulting children and young people, parents and practitioners is needed before we can be clear about what works for children with particular physical conditions to help their mental health, and why.

Word count: 294 words

Scientific Summary

Background

Having a long-term physical condition (LTC) places a strain on a child or young person (CYP) and their family. Having an LTC can adversely affect the mental health and wellbeing of CYP, which can in turn affect aspects of the physical condition such as treatment adherence and symptom severity. The overall risk of diagnosed mental health difficulties is reportedly around four times greater in CYP with LTCs than in their physically healthy counterparts. Although an extensive evidence base informs guidelines for the treatment of mental health disorders in CYP generally, there is a lack of evidence focusing on the treatment of mental health in CYP with long-term physical conditions. Therefore we aimed to address this gap by conducting two linked evidence syntheses and integrating the findings from these two reviews in an Overarching Synthesis.

Objectives

Two systematic reviews and an Overarching Synthesis of these reviews were conducted.

Review 1 aimed to evaluate the effectiveness and cost-effectiveness of interventions aiming to improve the mental health of CYP with LTCs and elevated symptoms of mental ill health.

Review 2 aimed to explore the factors that may enhance or limit the delivery of interventions aiming to improve the mental health and wellbeing of CYP with LTCs.

The Overarching Synthesis aimed to integrate the findings from Review 1 and Review 2 using a deductive approach.

The project aimed to integrate end-user involvement throughout the project in the form of input and feedback from topic experts, CYP with LTCs and their parents, as well as consultation on preliminary findings with a range of interested parties.

Summary of Review 1

Methods

Thirteen electronic databases were searched: MEDLINE, EMBASE, PsycINFO, Cochrane Database of Systematic Reviews, CENTRAL, DARE, HTA database, NHS EED, CINAHL, British Nursing Index, HMIC, Conference Proceedings Citation Index and Science Citation Index. This was supplemented with forward and backwards citation chasing, searches for sibling articles, website searching, author contact and searches for grey literature.

Two independent reviewers were involved in study selection, data extraction and quality appraisal. The inclusion criteria specified randomised controlled trials (RCTs) or economic evaluations involving CYP aged 0-25 years with LTCs and symptoms of mental ill health. LTCs were defined as any diagnosed physical health condition, with an expected duration of at least three months, where cure is considered unlikely, causing limitations in ordinary activities and necessitating medical care, or related services, beyond what is usual for age in question. Participants needed to have received an intervention that targeted their mental health, but there was no restriction on the type of intervention. Effectiveness had to be measured in terms of impact on at least one measure of the CYP's mental health.

Interventions and outcomes of included studies were categorised. Effect sizes for each study were calculated post-intervention using Cohen's *d*. Where multiple studies shared the same intervention type, similar comparator, similar participant LTC and the same outcome category, random effects meta-analysis models were fitted to pool effect sizes (*d*) across the studies.

Findings

A total of 25 RCTs (31 articles) that assessed the effectiveness of interventions aiming to improve the mental health of CYP with LTC and elevated symptoms of mental ill health were synthesised. These studies evaluated 11 types of intervention, sampling CYP with 12 different types of LTC. Outcomes were organised into 28 categories, 17 of which related to CYP mental health, the rest categorised as "other outcomes". Of the 11 reported interventions, the greatest volume of research focussed on the effectiveness of cognitive behavioural therapy (CBT), with ten papers (reporting on seven studies) evaluating this type of intervention. These studies provide tentative evidence that CBT-based interventions could be beneficial for the mental health of CYP with for the mental health of CYP with IBD, chronic pain, epilepsy and

persistent functional somatic complaints, but not type 1 diabetes mellitus. Some of the effective CBT interventions featured content which was adapted to the specific LTC prior to intervention delivery. Four parenting programme interventions were evaluated in three studies. Group play therapy interventions were also assessed in three studies. Other intervention types were seen in only one or two studies. Trials were typically small, meaning effect sizes across the included studies were characterised by wide confidence intervals. Therefore the evidence for particular interventions used with similar samples of CYP is very limited. There were relatively few opportunities for meta-analysis.

Review 1 highlighted the lack of trials aiming to address the issue of mental ill health in CYP with LTCs. In particular there were no relevant trials in the UK. The quality of the relatively small amount of studies was generally poor. The quality of future research in the field should therefore be improved. Although findings from Review 1 are tentative, there was some support for the findings from previous research.

The findings of Review 1 point to the need for large, high quality trials with consistency in intervention, design and outcome reporting. The development of manualised interventions would allow for rigorous testing of interventions across a range of conditions and locations and would aid comparison of the components of different interventions. It would be particularly useful to examine whether broad intervention types are effective across a range of long term conditions, including whether (and how) they should be adapted to specific LTCs. Although we included studies of interventions aiming to improve mental health, the majority also targeted other outcomes, particularly LTC symptoms. Therefore future research might consider integrated treatment and its effects across a wider range of outcomes, rather than focus primarily on mental health in terms of population, intervention and outcome.

Summary of Review 2

Methods

The databases MEDLINE (including MEDLINE in-process), PsycINFO, and CINAHL were searched. This was supplemented with forward and backwards citation chasing, searches for sibling articles, author contact, website searching and searches for grey literature.

Two independent reviewers were involved in study selection, data extraction and quality appraisal. The inclusion criteria specified primary qualitative studies that explore attitudes and

experiences regarding interventions aiming to improve the mental health and wellbeing of CYP with LTCs from the perspectives of CYP, their families and/or practitioners. Data analysis and synthesis broadly followed the principles of meta-ethnography.

Findings

In Review 2, 57 studies were synthesised, evaluating 21 types of intervention, a broader range than seen in Review 1. The most frequently seen LTC in the 57 included studies was cancer. Samples who were HIV positive or had a mix of LTCs were frequently seen, which differs from Review 1. Included studies commonly explored the perceptions and experiences of interventions aimed at improving coping, self-esteem and emotional support rather than targeting symptoms directly related to a mental health disorder such as depression or anxiety. Interventions often aimed to improve symptoms related to the LTC and social skills as well as mental health constructs. Included studies represented views from a range of different participants involved in the delivery and receipt of relevant interventions.

We used meta-ethnography to synthesise the included studies. The synthesis was presented as five main constructs: 'A Therapeutic Foundation', 'Social Support', 'A Hopeful Alternative', 'Resilience' and 'Getting In and Staying In'. A number of themes contributed to each construct. We developed a line-of-argument that offers an explanatory model of the experience of interventions to improve the mental health of CYP with LTCs.

The model describes the process by which CYP with LTCs access and maintain engagement with a relevant mental health intervention. Some interventions helped CYP to acquire a sense of hope for the future and increase their resilience. Interventions were often perceived to be effective when they offered participants a safe space and social support. The relationship between these constructs is tentative, as other factors may impact the experience of interventions and participants attitudes.

The quality of included articles was generally good. At least ten high quality articles contributed to each theme in the synthesis. The general weaknesses in this literature included a failure to make the theoretical perspective of the author explicit, to adequately describe the context or setting of qualitative research and a lack of clarity in the description of interventions.

We believe this is the first attempt to examine the experiences of participants in interventions that aim to improve the mental health of CYP with LTCs. However, our synthesis shares features with some previous literature. We identified only one study in which qualitative

data collection and analysis occurred alongside an RCT; there is a need for this type of mixed methods evaluation to improve our understanding of how interventions are experienced and how this may link to effectiveness. Our synthesis suggests that availability, access and engagement with interventions are crucial and that physical and mental health difficulties can pose unique challenges. Other aspects of interventions such as social support, relationships with those who deliver interventions and hope for the future are considered important by the range of people who received and deliver these interventions.

Summary of the overarching synthesis

Methods

The overarching synthesis integrated the findings from Review 1 and Review 2. A deductive approach was used whereby questions based on the findings of each review were generated and used to interrogate the other review for information that could potentially inform the findings or explain gaps in the literature. Despite the different research questions, mental health inclusion criteria, methods of synthesis and types of interventions seen across the two systematic reviews, the Overarching Synthesis allowed us to raise a number of tentative implications.

Findings

Nine categories emerged from the analysis:

- Degree of Overlap Between the Two Reviews
- Availability of Up-to-date, Good Quality Research
- What Works for Whom?
- Adaptations to Interventions and Flexibility
- Accessibility and Delivery
- Stress and Coping
- Working with Family and Peers
- Therapeutic Relationships
- Holistic Approach

There was a limited amount of overlap between the reviews, particularly when focusing on interventions used for particular populations. Much of the research included in both reviews was published in the ten years prior to the review, but there was a relative lack of studies conducted in the UK, with no effectiveness studies identified. While there is some evidence of benefit for interventions in CYP with particular LTC populations, for example cognitive

behavioural therapy in populations with inflammatory bowel disease, neither the quantitative or qualitative evidence can go further to explain why this may be or suggest how intervention effectiveness may vary between individuals.

There is some evidence from both reviews that interventions responding to the needs of individuals, particularly in relation to their LTC, may be beneficial. An intervention's setting, use of technology and flexibility can impact perceived effectiveness and may warrant consideration as to how such issues impact effectiveness. Alongside symptoms of mental health disorder, the overarching synthesis indicated elements of interventions which may support CYP to manage stress and coping related to their long term condition. There was evidence from both reviews that interventions which include family, particularly parents, may be beneficial. There also appear to be more benefits than disadvantages when children and young people are able to meet peers with similar health needs as part of an intervention. There is an indication that relationships between CYP with LTCs and their therapists and peers are important. While both Review 1 and Review 2 include interventions that aim to improve mental health in some way, the majority of interventions also targeted other outcomes including LTC symptoms, social support, knowledge and self-management.

Discussion and conclusions

In Review 1 the lack of similarity in terms of the combination of interventions, LTC and outcomes studied, often meant a reliance on small trials for effectiveness findings. Little meta-analysis of similar trials and no moderator analyses could be performed. This limited the strength of conclusions that could be made about intervention effectiveness. Despite conducting an additional database search and targeted searches, no economic evaluations were located. There were no UK studies included in Review 1, therefore limiting the applicability of review findings.

None of the studies included in Review 2 sampled participants with a diagnosed mental health disorder and only two considered the current mental ill health of participants upon study entry. The number of children and young people who received the interventions who were experiencing active mental health difficulties is uncertain and this limits comparability with Review 1. The Overarching Synthesis was challenged by the different inclusion criteria across Review 1 and Review 2. This may have partly explained the different intervention types seen

in each review, which presented a challenge to comparison. Review 2 interventions more often focused on improving coping, stress and self-esteem than Review 1.

There is some tentative evidence that cognitive behavioural therapy may be beneficial in some of the LTC populations in which it has been evaluated. Further research on what specific methods are currently used within paediatric healthcare services to recognise the mental health needs of CYP with LTCs, and their effectiveness, would be useful in helping to plan future interventions to address this apparent need. The delivery of interventions in both reviews indicates that it may be possible to deliver mental health interventions outside of clinical mental-health settings. However the training and support needs of intervention deliverers would need to be considered alongside the resources required for setting up and sustaining the intervention. Given the importance of social support, relationships CYP establish with those who deliver interventions, family and peers seem an important component of interventions.

The findings of Review 1 suggest the need for further large, high quality, well reported RCTs. In particular, improved reporting of the methods used to randomise participants and achieve allocation concealment, as well as blinding of outcome assessors is required. Review 1 suggests it would be useful to examine whether broad intervention types are effective across a range of LTCs, including whether (and how) they could be adapted to specific long term conditions. Interventions aiming to improve the mental health and wellbeing of CYP with LTCs often take a holistic approach; future research might therefore focus on integrated physical and mental health treatment and its effects across a wide range of outcomes, rather than focus primarily on mental health in terms of population, intervention and outcome.

Study registration

This study is registered as PROSPERO CRD42011001716.

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Foreword

Watching a child suffer from a long-term health condition is awful. They may be in pain, feel ill and be unable to do the things that other young people do. They may need surgery and lengthy stays in hospital. They need medication and side effects can be a challenge all by themselves. Watching a child cope with all this, and then put their fist through their bedroom door in anger, or refuse to leave the house for weeks through social isolation, or be unable to sleep at night/eat properly due to anxiety, is heart-breaking.

Children and young people with long-term health conditions – be it cancer, brain injury, muscular dystrophy or any of a myriad of physical conditions that can't be cured, only managed – face enormous challenges. As well as their physical illness, many of these young people suffer from mental health problems as a consequence of their condition. This may be anxiety, depression, anger, social isolation, poor self-esteem - amongst many others. They may be four times more likely to suffer mental health problems, than their physically healthy peers.

It is vital that the mental health of these young people is treated, alongside their physical condition. There are a range of strategies currently being used, and this study aims to evaluate how effective different types of intervention are, in improving mental health.

The quality and accessibility of these strategies varies greatly. Those young people lucky enough to have accessed top quality mental health provision, may overcome their anxieties and worries completely. They may recover to become confident and happy once again. Others may still not be able to attend school, engage socially or live without dark thoughts, years after their physical condition began. There is no justification for this disparity. Every single child or young person, suffering from a long term health condition, must receive top-quality intervention to improve their mental health – it shouldn't be down to chance as to which technique or provider they are given. Some parents have been able to see their child's anguished mental state healed. Others, on a daily basis, don't know what they will find when they return home each day.

In looking at the effectiveness of the various strategies, it is hoped that excellent practice can be identified and used to improve all mental health provision for our young people with long-term physical conditions. They deserve nothing less.

Fiona Lockhart

Chapter 1 Background

Description of the health problem

Defining long-term physical conditions

A 2007 systematic review of definitions of chronic health conditions in childhood found 25 different definitions used, with relatively few appearing across multiple publications.[1] For the current project, the team drew from some of the most frequently cited definitions,[2-4] to define long-term physical health conditions (LTCs) as:

Diagnosed physical health conditions, with an expected duration of at least three months, where cure is considered unlikely, causing limitations in ordinary activities and necessitating medical care, or related services, beyond what is usual for age in question.

Aside from this the definition was inclusive, with conditions including structural or functional central nervous system disorders (e.g. acquired brain injury, epilepsy), disability (e.g. cerebral palsy, spina bifida) or those with unclear aetiology (e.g. chronic pain, inflammatory bowel disease). It is acknowledged that long-term physical conditions may be referred to as chronic illness/disease/conditions or complex/special health needs and that most often these terms include mental health conditions, in the current review LTC refers to children and young people's physical health conditions only.

Defining mental health

The term "mental health" captures far more than the absence of psychiatric disorders,[5] to include adjustment, wellbeing and coping.[6] While we take this view of mental health, for the purposes of this project we needed to define mental health in different ways. When we refer to mental health conditions or psychiatric illness, we use the term "mental health disorder", for instance depression or conduct disorder. Since the reviews conducted do not focus only on mental health disorder, we use the term "mental ill health" to refer to elevated symptoms that may or may not relate to a particular mental health disorder, but indicate difficulties experienced by an individual, for example anxiety or stress. Finally we use the term "mental health and wellbeing" to refer to mental health in its broad sense as indicated above, to include coping and adjustment. This is necessary when we have been inclusive in terms of how

interventions may aim to improve mental health and the wide range of mental health outcomes that such interventions may affect.

Prevalence

Despite advances in medicine leading to improved prognosis and/or cures for many conditions, LTCs continue to be common in children and young people (CYP). Van der Lee and colleagues considered a range of definitions of chronic conditions, but identified an overall prevalence of around 15% in CYP in the international literature.[1] In the USA, prevalence of LTCs is high and has increased over time. The U.S. National Health Interview Survey (NHIS) has annually collated information about the health of CYP across the nation, and in 2014, 42% suffered from one or more LTCs (NHIS data, childhealthdata.org), while in 1988 this number stood at 31%.[7] Although the majority of reported data relates to the U.S., a high prevalence of LTCs is experienced across the globe. In England, 23% of secondary school age pupils reported that they had a long-term medical illness or disability,[8] with similar proportions of New Zealand, Canadian and Finnish youths reporting an LTC.[9, 10] Inconsistency in the methods used to survey the health of CYP exist, for example in the definition of LTCs, the sample inclusion criteria, or the reporting method; in spite of uncertainty regarding the prevalence of LTC according to our definition, there is strong evidence that there is an international burden related to LTCs in CYP.[11]

Having an LTC places strain on the individual and their family, as well as placing demands on societal systems such as healthcare and education. A mental health disorder in addition to the LTC therefore poses a significant problem for some CYP and those around them. The challenges associated with childhood mental disorder alone can be significant and have the potential to exert a more negative impact than certain LTCs.[12]

There is extensive evidence that links the presence of an LTC with increased risk of the development of a mental health diagnosis in CYP. Although the overall risk of mental health disorder is reportedly around four times greater in CYP with LTCs than in their physically healthy counterparts,[13, 14] this risk varies due to a number of factors. For example, the duration[9, 15, 16] severity or progression[10, 17, 18] of an LTC are associated with mental ill health or risk of mental health disorder.

For a variety of LTCs, there is evidence to support increased risk of mental health disorder: for example asthma;[19, 20] gastrointestinal disorders;[21] functional abdominal pain;[22]

kidney disease;[16, 23, 24] chronic headache;[25] type 1 diabetes mellitus[26] and epilepsy,[27]. However, for a range of LTCs there is limited evidence of any association with mental health disorder and many studies suffer from poor quality. For example, the literature regarding CYP with cancer is particularly equivocal. There is research showing little difference in depressive symptoms between CYP with cancer and healthy comparators (e.g. [28-30]), although aspects such as the stage of treatment,[31] or undertaking certain distressing procedures, such as stem cell transplants,[32, 33] can influence the mental health of cancer patients.

How long-term physical conditions may increase risk of mental ill health

While the prevalence of mental health disorder in populations with LTCs may be greater than populations without such physical health concerns, the mechanisms for this are not fully understood. Physical and mental health conditions can share the same pathology,[34] or the stress and/or treatment associated with having an LTC can adversely affect mental health. For instance, LTCs and their treatment may involve: pain, invasive, complicated or time-consuming treatment regimens, time away from school and peers for hospital visits; restrictions on activity or diet, and a sense of isolation from friends as well as family.[35]

There are several empirical models which attempt to explain why children with an LTC are at risk of mental ill health, including the Risk-Resistance Model developed by Wallander and Varni.[36, 37] This model was intended to guide interventions to support children to adapt to living with an LTC. It suggests that aspects of disease severity, the impact an LTC has on the child's functional independence and other psychosocial stressors within the child's life are a set of risk factors which can increase the risk of a child developing some form of mental, social or physical maladjustment associated with their LTC. The model also proposes a set of personal and family factors which may act as factors protecting against mental ill health. Both risk and protective factors identified by this model are proposed to be applicable across different types of LTC, but their relevance to specific illness groups has not been widely tested.

An alternative theory is Thompson's transactional stress and coping model,[38] which views chronic illness using a systems theory approach and sees an LTC as a stressor requiring adaptation. This model identifies processes that contribute to the adjustment of children with chronic disorders and their mothers. Adaptation to an LTC is seen to involve psychological, biomedical and developmental processes and therefore there are similarities to Wallander and Varni's model.[37] Thompson's model has been tested primarily with sickle cell disease and cystic fibrosis.[39]

Moos and Holahan's conceptual model for the Determinants of Health-Related Outcomes of Chronic Illness and Disability[40] builds on the previous models by integrating the influence of personal and social factors on the course of illness. It demonstrates how different adaptive tasks can influence development of coping skills and mediate potential health outcomes. The model emphasises the inter-relationship between both risk and protective factors and health-related outcomes, resulting in the concept of adjustment to an LTC as a process rather than an endpoint. Whilst aimed at adults and thus omitting the important influences of developmental stage and family and social dynamics for CYP, the model notes the importance of building relationships with healthcare providers as one of seven "adaptive tasks" for the chronically ill person to complete.

Haase and colleagues put forward a Resilience in Illness Model, which focuses on adolescents and young adults, identifying risk and protective factors that influence the resilience shown by those experiencing illness-related distress.[41] In common with the above models, family factors and coping are considered, which has had exploratory and confirmatory evaluations amongst young people with cancer.

Aside from theoretical models of the interaction between physical and mental ill health, aspects of the experience of living with an LTC highlight the stress that CYP face. The initial diagnosis of an LTC is often very distressing for both the child and their families and associated with feelings of shock, sadness and confusion.[42] At the time of diagnosis, children and families must take in information regarding the child's immediate treatment needs which can feel confusing and frustrating, but also consider the possible impact of the diagnosis on the child's future.[43]

The daily experience of living with an LTC can be physically unpleasant for CYP, and this may have a detrimental effect on their mental health via several mechanisms. Venning and colleagues conducted a qualitative meta-synthesis, focusing on the experiences of children diagnosed with an LTC and noted how LTCs made children "feel uncomfortable in their body and the world"[44](p. 329).

Impact of mental health on physical health and economic consequences

Cottrell[45] describes the vicious cycle of comorbid LTC and mental health diagnosis, where one condition exacerbates the other. Mental ill health may also be associated with poor treatment adherence, which may exacerbate the physical condition, impair self-management

and worsen long-term outcomes.[46, 47] For example, depression in children with diabetes is associated with poorer control of blood sugar, increasing the risk for later serious complications such as loss of vision.[48] It is estimated that between 12 and 18 per cent of all NHS spending on LTCs is linked to poor mental health and wellbeing[49] and that psychological interventions can reduce care costs by up to 20 per cent.[50]

Interventions

There are numerous highly-effective psychological interventions for mental ill health in CYP, where there has been no explicit regard to physical illnesses, with over 750 treatment protocols cited in one systematic review.[51] For instance, the National Institute for Health and Care Excellence (NICE) recommends the use of group or individual parenting interventions for children with conduct disorder,[52] and cognitive behavioural therapy (CBT) for depression.[53] It is important to know whether such interventions are also effective in children with physical illnesses. However, children with some LTCs are often excluded from trials of such interventions; for example, in a large trial of CBT for children with anxiety disorders, children with a disabling medical condition were excluded.[54]

Various characteristics of an LTC may make traditional interventions for mental ill health more challenging and/or less effective, requiring a modified approach to treatment. Cottrell[45] suggests that while existing evidence-based treatments for mental health diagnoses in CYP should be effective in the presence of LTCs, there are circumstances where this may not be the case. For example, in CYP with type 1 diabetes, the strong link between blood glucose control and mood disorders[55] may mean that management of diabetes should be targeted first.[56] In CYP with epilepsy where seizure control is challenging, it is recommended that any treatment for depression is carefully managed by an interdisciplinary team and adapted to individual cases, particularly where pharmacological management is warranted for either condition.[57] Patients with comorbid gastrointestinal disorders and depression may be unsuitable candidates for pharmacological treatments, as they may exacerbate physical symptoms, while approaches such as cognitive behavioural therapy may need to be adapted to focus on maladaptive thinking related to the LTC.[58]The additional costs of treatment for mental ill health can be significant. For example, the presence of comorbid depression in adolescents with asthma in the USA is suggested to increase the costs associated with treatment by 51 per cent, compared to those with asthma alone.[59]

Treatment guidance

The closer integration of mental and physical healthcare is a priority for the NHS[60] and the NHS Confederation has highlighted the social, health and economic benefits that arise from integration of physical and mental health treatments.[61] NICE calls for access to mental health professionals with an understanding of diabetes to address psychological and social issues in CYP with diabetes[62] and state that the psychological needs of children with epilepsy should be considered as part of routine care.[63] A 2014 Department of Health policy paper[64] builds on the Government's mental health strategy for mental and physical health to have "parity of esteem"[60](p .2). It explicitly states its ambition for mental health care and physical health care to be better integrated at every level and mandates best practice approaches to caring for patients to include potential psychological care needs. However, at present we do not know what 'best practice' consists of in relation to the treatment of mental ill health in CYP with LTCs.

In 2015,[65] the Department of Health and NHS England released a policy document, 'Future In Mind', which aimed to comprehensively evaluate the current picture of children's mental health in the UK, and to outline plans to improve the care provided. Within this document, the prevalence of comorbid mental health diagnosis in CYP with LTCs is highlighted, but the associated treatment difficulties are not discussed and the only recommendations related to this topic are aimed at holistic school-based promotion of mental and physical self-care. NICE has not produced any general guidance for the treatment of CYP with LTCs and comorbid mental health diagnosis, although it exists for the treatment of adults with LTCs and depression.[66] One document specifically considers mental health services referral in diabetes patients,[62] but this is focused more on management of the physical condition. Specifically, NICE guideline NG18 acknowledges the increased risk of emotional and behavioural difficulties in CYP with type 1 diabetes, and promotes awareness of these risks in healthcare professionals. The availability of mental ill health screening and referral to mental health professionals with expertise in diabetes is recommended.

This represents a gap in policy development and research activity. In adults with cancer, the collaborative care model implemented by Sharpe and colleagues[67] led to large improvements in depressive symptoms compared with usual care. The model involved the integration of cancer nurses and psychiatrists into the hospital environment alongside primary

care physicians, and given the improvements shown in treatment of depression in the adult population, primary evidence is needed to assess such an approach with CYP with LTCs.

Evidence for adults cannot simply be applied to CYP. A wide range of factors may influence the effectiveness of interventions for CYP.[68] For example: the developmental stage of the child; aspects relating to parents; accessibility requirements, such as relying on others to access treatment; attentional requirements, such as keeping children engaged and interested; the need to work around education; different social aspects of being a child compared to being an adult. It is clear that separate research is required for CYP with LTCs.

Measurement of mental health and other outcomes

There are a range of different outcome measures that can be used to measure the effectiveness of mental health interventions for CYP with LTC. There are several reasons for this. Firstly, there is a wide range of mental health disorders that might co-exist with LTCs and likewise varied symptoms of mental ill health which could be measured using a large number of validated scales. Secondly there are no gold standard outcome measures. For example, a Cochrane review of CBT for anxiety in CYP that was restricted to studies using validated and reliable diagnostic interviews and symptom rating scales identified four diagnosis tools and more than eight different outcome measures in 41 studies.[69] Thirdly, some more generic measures of mental health and wellbeing are used to measure effectiveness. Finally, interventions which aim to improve the mental health of CYP might also be predicted to improve markers of physical health, whether as a result of improved mental health or components of the intervention.

Previous systematic reviews

Systematic reviews of mental health interventions rarely separately consider the effectiveness of these interventions in populations with LTCs. A Cochrane systematic review by James and colleagues included 41 studies to assess the effectiveness of CBT for anxiety in CYP.[69] Despite this relatively large number of studies, none considered a sample or subgroup where a comorbid LTC was present. Similarly, systematic reviews of non-pharmacological treatments for depression in CYP with traumatic brain injury[70] and congenital heart disease[71] found no relevant randomised controlled trials (RCTs) in CYP.

There are a number of existing, well-executed systematic reviews in the broader area of psychological interventions for people with LTCs, including several Cochrane reviews.[72-75]

However, these reviews are highly targeted; for example, focusing on one particular physical and/or one particular psychiatric comorbidity, not focused exclusively on CYP, or focused exclusively on parenting interventions.

The majority of other reviews in this area have concerned either adult populations or specific disorders, and have often focused on the distress arising from having an LTC rather than the treatment of a comorbid mental health disorder.[56, 76] Reviews of psychological interventions in CYP with LTC have been undertaken, but many of these have focused on coping, treatment adherence, and/or use of healthcare resources in populations without elevated mental ill health (e.g.[77-79]). There have been some reviews of pharmacological interventions for children with mental health diagnoses and physical illness, which cover single physical illnesses and mental health symptoms (e.g. depression in epilepsy[80]).

The most relevant attempt to systematically review psychological interventions to treat symptoms of mental health disorder in CYP with LTCs was published by Bennett and colleagues.[81] This review targeted only psychotherapeutic interventions, where child-related mental health measures were the primary outcome. A total of ten relevant studies were identified, of which two were RCTs. These RCTs trialled CBT for sub-threshold depression in CYP with epilepsy[82] and inflammatory bowel disease.[83] Although the review provides preliminary evidence that CBT can be an effective treatment for depression and anxiety in CYP with LTCs, reviewers concluded that the existing evidence base was weak.

The current review fills a gap in the literature by synthesising studies investigating interventions aiming to improve mental health where CYP with LTCs have elevated symptoms of mental ill health and studies reporting cost and cost-effectiveness evidence. In addition, this review synthesises evidence from qualitative research studies, in order to understand how children with LTCs experience interventions aimed at improving their mental health and wellbeing and their attitudes towards them.

Aim and research questions

We aimed to evaluate the effectiveness of interventions aimed at improving the mental health of children and young people with long term physical health conditions and to explore the factors that may enhance or limit the delivery of such interventions. This necessitated reviewing both quantitative and qualitative research. The systematic reviews addressed the following research questions:

1) What is the effectiveness and cost-effectiveness of interventions targeting mental health for CYP with LTCs and symptoms of mental ill health?

2) What are the effects of such interventions on other key aspects of individual and family functioning?

3) What are the factors that may enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions intended to improve mental health for CYP with LTCs?

Chapter 2 Review 1: Effectiveness of interventions aiming to improve mental health in children and young people with long term physical conditions

Research questions

This chapter describes the first systematic review and addresses the following research questions:

1) What is the effectiveness and cost-effectiveness of interventions aiming to improve mental health for children and young people (CYP) with long term conditions (LTCs) and symptoms of mental ill health?

2) What are the effects of such interventions on other key aspects of individual and family functioning?

Methods

The methods used to identify and select evidence followed recommended best practice guidance.[84, 85] A protocol for the systematic reviews across the project was registered on the PROSPERO database (PROSPERO CRD*******).

Identification of evidence

Inclusion/exclusion criteria

The following inclusion and exclusion criteria were used to determine eligibility of articles. The inclusion criteria specified randomised controlled trials or economic evaluations involving CYP aged 0-25 years with LTCs and symptoms of mental ill health. Participants needed to have received any type of intervention that targeted their mental health. Effectiveness had to be measured in terms of impact on at least one measure of the child or young person's mental health. Further details of inclusion and exclusion criteria are shown in *Supplementary Materials 1,* .

Search strategy

A search strategy was developed and tested in the databases to be searched. The strategy used both controlled headings (e.g. MeSH) and free-text searching. Terms were grouped according to four concepts:

- Children and young people terms
- Mental health terms
- Long term physical conditions terms
- Study design terms (using Cochrane filter for locating randomised controlled trials[86])

LTC terms were informed by previous reviews including studies with populations with LTCs or chronic conditions,[78, 81, 87-104] as well as discussion with experts amongst the wider project team. Thirteen electronic databases were searched: MEDLINE including MEDLINE in-process (via OvidSp), EMBASE (via OvidSp), PsycINFO (via OvidSp), Cochrane Database of Systematic Reviews (via the Cochrane Library), CENTRAL (via the Cochrane Library), DARE (via the Cochrane Library), HTA database (via the Cochrane Library), NHS EED (via the Cochrane Library), CINAHL (via EBSCOhost), British Nursing Index (via ProQuest), HMIC (via OvidSp), Conference Proceedings Citation Index (via Web of Science), Science Citation Index (via Web of Science). No language or date restrictions were applied. Searches were conducted between 28th January and 4th February 2016. An example search strategy used for the MEDLINE database is shown in *Appendix 1*. All references identified by the searches

were exported into EndNote X7 (Thomson Reuters, New York, USA) prior to de-duplication and screening.

A second search strategy was designed to locate studies relating to cost-effectiveness using the University of York's Centre for Reviews and Dissemination (CRD) search strategy for economic evaluations in place of the randomised controlled trials filter.[105] The economic evaluation filter for this search was applied to MEDLINE and EMBASE from April 2015 only as the NHS Economic Evaluation Database (EED) was updated using these databases up until 31st March 2015. Searches for economic evaluations were carried out on 3rd May 2016. The search strategy used for EMBASE to locate economic evaluations is shown in *Appendix 2*.

Supplementary searches were also conducted. Backward citation chasing (searching the references of included articles) w conducted by three researchers (DM, MN, LS) to locate further potentially relevant articles. Alongside backward citation chasing, these researchers checked lists of included studies from related reviews. Forward citation chasing (searching articles citing included articles) was conducted by an information specialist (JTB) using Web of Science and Google Scholar. In addition, DM, LS and MN searched websites that had been identified by the project team and children and young people advisory group for relevant research (see *Appendix 3* for a list of websites searched). Targeted searches to identify "sibling" papers (further outcomes, process evaluations, economic studies and qualitative research) associated with included trials and based on trial names and first and last authors were conducted by JTB. MN emailed all contact authors of included studies to request any articles associated with included articles. The databases CINAHL, HMIC and Conference Proceedings Citation Index were searched, all of which index grey literature. The website OpenGrey was also searched via http://www.opengrey.eu/ on 23rd June 2016.

Study selection

Relevant studies were identified in two stages based on the inclusion/exclusion criteria given above. First independent double screening of titles and abstracts for each record was conducted (seven researchers shared this screening: MN, LS, DM, JTC, MR, VB, IR). Endnote X7 was used to perform screening. Disagreements were resolved through discussion between two reviewers, with referral to a third reviewer as necessary (DM, MN, LS). Full texts of records that appeared to meet the inclusion criteria on the basis of titles and abstracts were then obtained wherever possible via the University of Exeter online library, web searching and the British Library. Each full text article was screened independently by two reviewers (six

researchers shared this screening: MN, LS, DM, JTC, VB, IR). Reasons for exclusion at this stage were recorded. Disagreements were again resolved as for title and abstract screening. . To assess population eligibility at full text screening, reviewers often needed to locate information regarding cut-off scores for validated measures of mental health outcomes. These were rarely reported in screened records and therefore reviewers searched for publications including cut-off scores for these measures or manuals for the scale in question. Wherever possible a threshold indicating symptoms of clinical distress in a previous CYP sample was used. These thresholds were collated so reviewers applied them consistently. Screening for the additional economic search proceeded in the same manner.

Methods of analysis/synthesis

Data extraction

A data extraction form was developed and piloted. MN, LS and DM each extracted one article and checked one colleague's extraction, before discussing and amending the form. Data on article details and aims, participants, mental health measures at baseline, intervention, outcome measures, findings and study quality were extracted into Microsoft Office Excel 2010 (Microsoft, Redmond, CA, USA) by MN, DM and LS and checked by either DM or MN. Where data were missing that would have allowed for meta-analysis, authors were contacted for information alongside our request for sibling papers.

Quality appraisal

Quality appraisal was conducted simultaneously with data extraction using criteria adapted from the Cochrane risk of bias tool.[85] In addition to criteria on randomisation, allocation concealment, blinding of assessors, and selective outcome reporting from the existing tool, we included items on intention-to-treat analysis, between-group similarities at baseline, dropouts, response rate, intervention detail, intervention manuals, adherence, follow-up measures and psychometric properties of outcome measures (c.f. Richardson et al.[106]). This gave 15 items on which risk of bias and quality of included articles were assessed, providing additional insight into study quality and reporting. Assessment of quality and risk of bias was performed at the article level, in order to evaluate any differences on account of the different outcomes reported in articles from the same study. Quality appraisal decisions were made by two reviewers (from DM, MN and LS) and disagreements resolved through discussion. The

appraisals were used to evaluate risk of bias and study quality and were not used to exclude papers.

Categorisation of interventions and outcomes

Intervention categories

During data extraction, interventions were categorised according to similarities in terms of broad intervention type and intervention content. The label and definition of these intervention categories were developed using the descriptions of interventions in the included studies and with reference to previous general classifications of interventions. The categories were developed by DM and MN and discussed with the wider team. The categorisation was primarily used to organise the presentation of the synthesis by intervention type, with sections synthesising the findings from studies relating to each intervention category.

Outcome categories

Due to the diversity of outcomes used to measure the effectiveness of interventions in the included studies, outcome categories were determined in response to the constructs measured by the instruments used. These categories, and therefore each measure, were also categorised at a broad level as either a 'CYP mental health outcome' or 'other outcome', given that the focus of the review is upon interventions that aim to improve CYP mental health. Outcomes were categorised as CYP mental health when they appeared to meet our broad definitions of mental health and wellbeing given in *Chapter 1*. Other outcomes were extracted and analysed in order to include all effectiveness outcomes and therefore address our second research question (What are the effects of such interventions on other key aspects of individual and family functioning?). These categories and the measures used within included studies are shown in the tables in the *Analysis of included study findings* section of this chapter. These outcome categories were refined after data extraction by MN and then reduced by DM, before sharing with experts amongst the wider team. These outcome categories were primarily used to determine where results for outcomes measuring similar constructs could be meta-analysed and as a way to compare similar outcomes for each intervention category, without necessitating knowledge of individual measures.

Data analysis and synthesis

The principal summary measure used to compare effectiveness findings in included studies was differences in means between intervention and control group at post-test and, where

available, the longest follow-up time point. For each outcome, means, standard deviations (SDs) and sample sizes (or figures that could be used to derive these where available) for the relevant intervention and control groups were used to assess differences between groups. Where we did not receive necessary data for outcomes either from articles or correspondence with authors we did not synthesise these outcomes. Where such figures were not available for any CYP mental health outcomes in a study we synthesised findings narratively. Effect sizes for each study were calculated post-intervention using Cohen's *d*; that is, the difference between the means in the two groups divided by their pooled standard deviation.[107] Ninety-five per cent confidence intervals for the effect sizes were also calculated, along with *p*-values. Effect sizes and confidence intervals were calculated using the *metan* command in Stata software (StataCorp, College Station, TX, USA). *P*-values for each effect size were calculated using the *ttesti* command in Stata. Cohen's guidelines were used to aid interpretation of effect sizes.[107] Classifications for what are considered to be 'small', 'medium' and 'large' effect sizes are at least $d = 0.20$, $d = 0.50$ and $d = 0.80$, respectively. While these classifications are widely used, it is acknowledged that these guidelines do not take account of how the clinical or practical importance of effects might vary between outcome measures.[108]

Where multiple studies shared the same intervention type, similar comparator, similar participant LTC and same outcome category, meta-analysis was considered feasible. Random effects meta-analysis models were fitted to pool effect sizes across the studies, based on the assumption that no two studies were addressing the same research questions in exactly the same way. For each pooled effect size estimate, we calculated 95% confidence intervals. The I^2 statistic (possible range 0% to 100%) was used to quantify statistical heterogeneity with higher values indicating greater heterogeneity.[109] When two or more measures assessing the same outcome category were reported in a study, the effects were combined into a single summary effect for that study, calculating the standard error for this effect using the correlation between the measures, obtained from the paper itself or other research.[110] When different studies used identical measures we conducted additional meta-analysis using raw mean differences. All meta-analyses and associated forest plots were produced using the *metan* command in Stata.

We intended to assess publication bias by examining funnel plots for asymmetry using the *metafunnel* command in Stata. However, we were unable to assess funnel plots properly or use more advanced regression-based assessments to assess publication bias owing to the substantial heterogeneity identified across studies and small number of studies with similar

characteristics that could be entered into a given meta-analysis.[111] Therefore, we cannot comment on publication bias in this review.

Results

Tables and Figures described below are available in Supplementary Materials 1.

Study selection

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) diagram[112] in *Figure 1* summarises the process of study selection. Around five per cent of the 18,360 records were identified by means other than searches of academic databases, including citation chasing searches, searching relevant reviews and websites, searches for sibling papers and author contact. After the removal of duplicates, a total of 11,706 records were screened at title and abstract stage. The full texts of 343 records were retrieved for further consideration. Of these, 336 (98 per cent) were successfully retrieved. After full text screening 312 articles were excluded for reasons provided in *Figure 1*. The majority of articles were excluded because CYP mental health measures did not indicate that the sample was above an established cut-off at baseline, no recognised cut-off was available for the measures used, or there was not enough information to determine inclusion because only an abstract had been published. A number of articles were excluded because they did not meet inclusion criteria for one of intervention target, population or study design. Only one paper was excluded on account of the sample having moderate intellectual disabilities. A list of reasons for the exclusion of each article screened at full text is located in *Supplementary Materials 1, Table 2*.

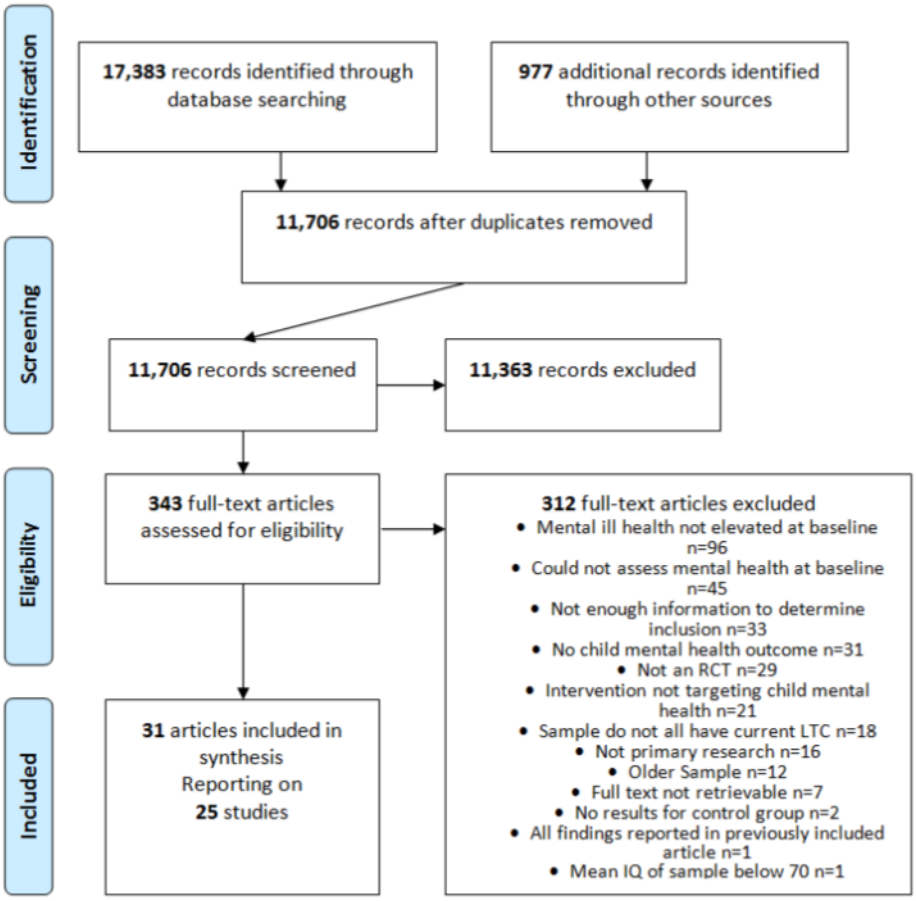

Figure 1: PRISMA flow chart recording Review 1 study selection.

Thirty-one articles met our inclusion criteria and were included in the synthesis. Six of the articles were additional papers for studies already reported in included papers. These papers were included if they provided any additional relevant data not seen in other study articles. Eight of the 31 included articles were located through additional searching (forward citation chasing, backward citation chasing, author contact, included in previous reviews). Twenty-two authors from the 25 included studies were contacted via email requesting details about any other articles associated with the studies. Ten authors responded. Specific data queries were included in ten of these emails. Six authors replied, allowing additional effect sizes to be calculated. Of the 31 articles that met the full-text screening criteria, there were no economic outcomes such as costs or cost-effectiveness reported.

Economic evaluation

The PRISMA diagram in *Figure 2* summarises study selection after the second database search for economic evaluations and targeted search for sibling studies of the 31 articles included in the review. The majority (78 per cent) of the 207 records identified were from the

search of databases. Thirteen records were considered relevant after title and abstract screening. Both reviewers agreed that none of the thirteen articles met the inclusion criteria for this part of the review. Of note amongst the screened articles were four studies. Whittemore and colleagues[113] provided a single cost estimate of providing a five-week Teencope intervention that met the intervention inclusion criteria, but there was no breakdown of this cost or description of how it was calculated. Figures in another study[114] on using 'integrated psychological therapies' to increase treatment adherence (the Shine intervention), allowed estimation of the cost of providing the intervention/therapy to 12 patients as £4,260. However, some costs (such as hospital staff training and costs of staff from the social enterprise) appear to have been excluded, and there were insufficient data presented for the stated net cost savings to be seen as reliable.

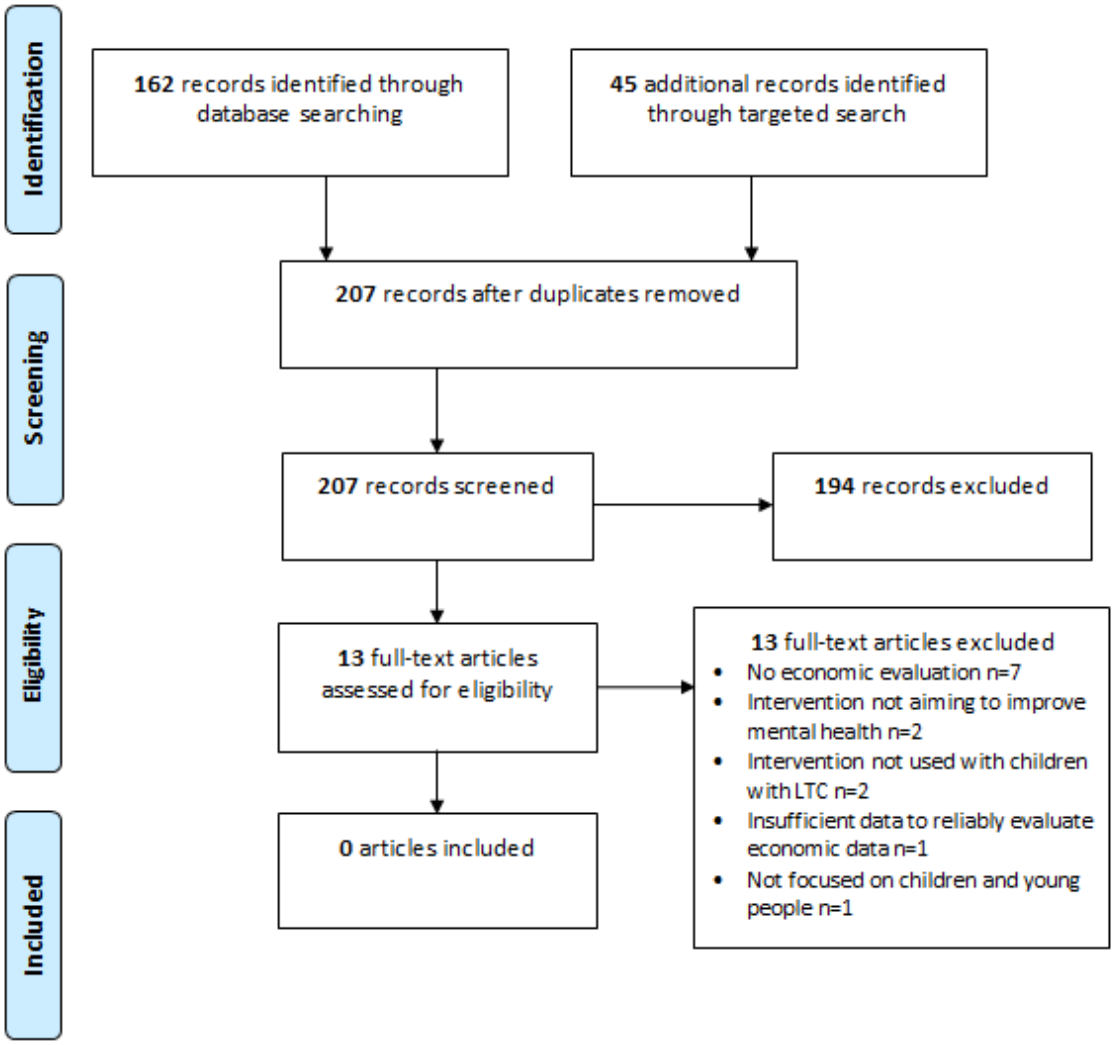

Figure 2: PRISMA flow chart recording Review 1 economic evaluation study selection.

It was noted that two of the screened economic studies assessed the cost of the Triple P System parenting intervention (Foster et al.[115] in USA; Mihalopoulos et al.[116] in Australia) which was also evaluated in one of our included effectiveness studies for Review 1.[117] However, there were different programme features between the intervention used in the study by Westrupp and colleagues and the other two studies (which would have made the costs different) and both the studies screened here targeted all families with at least one child under particular ages; so they did not specifically target families with children with LTCs. For both these reasons, their findings would not be generalisable to either Westrupp and colleagues[117] or similar parenting interventions which only targeted families with children with LTCs. They were therefore also excluded.

Descriptive statistics

Supplementary Materials 1, 3 provides details about the study location, participants' LTC, intervention type, comparator, qualifying baseline mental health status and other reported CYP mental health outcomes for the 25 studies included in this review. Studies were conducted in eight different countries, with the USA (n = 10:[83, 118-126]) most common, followed by Australia (n = 5:[117, 127-130]) and Iran (n = 4:[131-134]). Studies included children or young people with 12 different LTCs: cancers (n = 5:[122, 132, 134-136]) receiving the most attention. Type 1 diabetes mellitus (T1DM) (n = 4:[117, 119, 121, 129]), asthma (n = 3:[118, 137, 138]), inflammatory bowel disease (IBD) (n = 3:[83, 124, 125]) and hearing loss (n = 2[131],[133]) were also the subject of multiple studies, with all other LTCs featuring in one study each. Only one article was published before 2000,[119] with 13 articles (52 per cent) being published since 2010.[117, 118, 122-127, 129-133, 136, 137]

Five included studies were reported in more than one included journal article reporting on different study details or findings. Brown and colleagues' 2015 paper[139] reported additional parent and family function outcomes from their 2014 trial.[127] We excluded one article associated with this study as it did not provide any effectiveness outcomes that were not seen in the previous articles.[127] Lyon and colleagues included detailed information about the content of their intervention in 2013,[140] with outcome data reported in the 2014 article.[122] The earliest Szigethy and colleagues[83] trial was linked with two additional papers. These reported on correlates of treatment with individual items on the Child Depression Inventory (CDI)[141] and long-term follow-up data for the original trial.[142] For Szigethy and colleagues' later trial, the 2015 article[143] reported secondary outcomes from a subset (those with Crohn's disease) of the 2014 sample, which included both Crohn's Disease and Ulcerative Colitis patients.[125] Finally, the two papers from Whittingham and colleagues reported different outcomes from their trial of a parenting and acceptance and commitment therapy (ACT) for children with Cerebral Palsy.[130, 144] The 2014 paper[50] focused on directly on child behaviour outcomes and parenting skills, while the 2015 paper[64] reported child quality of life related to physical functioning, and further parenting and parent mental health outcomes.

A range of interventions were evaluated, with those categorised as cognitive behavioural therapy (CBT) the most frequent (n = 7:[82, 83, 123-125, 129, 145]). After CBT, interventions categorised as parenting programmes and group play therapy were most commonly evaluated. Emotional intelligence training,[131, 133] stress management programmes[119, 121] and palliative care[122, 136] also featured in two studies each. Most often, the comparator involved 'usual care'

(e.g. regular schooling, usual sedation, usual hospital care) within a given setting (n = 19:[82, 83, 117, 119, 121-123, 126, 127, 129-136, 138, 145]), with five of these studies employing a waitlist control group.[121, 123, 136, 126, 130] Active comparators in the remaining six studies included asthma education,[118] progressive muscle relaxation,[120] aerobic exercise[128] and non-directive supportive therapy.[124, 125]

Two studies contained in their inclusion criteria a mental health diagnosis (principal anxiety disorder in Masia Warner and colleagues;[123] mild or major depression in Szigethy and colleagues.[125] All other studies included samples where reviewers found that symptoms of mental ill health were above an established threshold or included a sample at risk for a mental health disorder.[82] At baseline, elevated symptoms of anxiety[118, 120-124, 126, 135, 136, 138] and depression[82, 83, 120, 125, 128, 132, 136, 145] were most common, each present in ten studies. The participants in seven studies showed elevated symptoms in more than one mental health domain.[83, 120, 123, 131, 133, 136, 138] Additional CYP mental health outcomes were reported in 15 studies, crossing a wide range of categories including anxiety and depression, coping measures, emotional difficulties, adjustment and general mental health.

Sample characteristics

The studies included a total of 1198 participants, of whom 48.1 per cent were female, with a mean age of 12.2 years (where data were available: gender not reported by Gordon and colleagues[128]; mean age not reported by Hains and colleagues[121] and Zareapour and colleagues[134]). The ethnicity of the sample was predominantly (>66 per cent) white in seven studies[83, 119, 121, 123-125, 127] and predominantly black or African American in two studies.[118, 120] Lyon and colleagues[122] and Yetwin[126] recruited samples from a more diverse mix of ethnicities, but ethnicity was not reported in 12 studies. *Supplementary Materials 1,* provides details about the sample size, gender, age, ethnicity and inclusion criteria for each study.

Eleven studies provided some information about the socioeconomic status of participants.[83, 117, 119, 120, 122-125, 127, 130, 136] The method of classifying socioeconomic status was inconsistent, with family income,[83, 123-125, 127, 130] federal poverty level,[122] parental education,[83, 122, 127, 130] parental employment status,[127] social class,[136] a socioeconomic index[117] and the Hollingshead Index[119, 120] all reported, preventing comparison across the included studies.

Thirteen studies recruited subjects from hospitals.[83, 117, 119-122, 125, 129, 132, 134-136, 145] Referrals by specialists or general practitioners, with or without additional advertising in clinics or waiting rooms (flyers, online posts etc.) was the second most common method of recruitment to studies, used in six studies.[82, 123, 124, 126-128] Three further studies recruited from schools[118, 131, 133] and two by consulting patient databases.[130, 138] In total 111 (9.2%) participants dropped out from studies after the intervention delivery had commenced, which appears low.[146]

Typically, inclusion criteria in included studies required diagnosis of the relevant LTC, lack of intellectual disability that would prevent understanding of interventions or assessments, and willingness/availability to participate. Five studies had specific requirements of elevated mental health symptoms for inclusion. Martinović and colleagues[82] required 'subthreshold' depression scores (just below a validated cut-point on three scales) while Szigethy and colleagues[83] required a minimum score of nine on the Children's Depression Inventory. In the 2014 study by Szigethy and colleagues[125] diagnosis of minor or major depression was required and confirmed using the Kiddie-Schedule for Affective Disorders and Schizophrenia-Present Version (K-SADS-PL) diagnostic interview. Brown and colleagues[127] included parents who subjectively considered their child to have behavioural problems. Masia Warner and colleagues[123] recruited participants with a previously diagnosed principal anxiety disorder. However, 13 studies excluded participants with certain mental health diagnoses or issues.[82, 83,

[117, 122-126, 129, 131, 134, 137, 138] Therefore some studies excluded particular mental health diagnoses, even when other diagnoses or elevated symptoms were required.

Intervention characteristics

Supplementary Materials 1, displays details about the interventions reported in the included studies. The intervention aims and structure, as well as details of the delivery site and personnel, the intended recipients and comparator group are shown. Eleven intervention categories were used to group together similar interventions. The label and definition of these intervention categories are shown, along with the studies evaluating them, in *Appendix 4*. Interventions based on CBT were the most frequently studied (n = 7:[82, 83, 123-125, 129, 145]), with n = 267 participants randomised to this type of intervention. The two studies reported by Szigethy and colleagues[83, 125] explored the effects of the CBT programme PASCET-PI (Primary and secondary control enhancement therapy-Physical illness) on various outcomes in children with inflammatory bowel disease,[83] or Crohn's disease specifically.[143] Masia Warner and colleagues[123] and Reigada and colleagues[124] used versions of TAPS (Treatment of Anxiety & Physical Symptoms), the latter modifying the intervention to include management of specific symptoms of inflammatory bowel disease. Adapted intervention content was seen in eight studies.[83, 118, 119, 123-125, 129, 145] All but two of these were trials of CBT, with Bignall and colleagues'[118] relaxation intervention and Boardway and colleagues'[119] stress management training also including adapted content. For each study, *Appendix 5* describes the content of each intervention, and whether it was adapted or delivered flexibly. Only Szigethy and colleagues specifically allowed flexibility in delivery,[125] allowing for individual changes to content based on the developmental stage of the recipient.

The parenting programmes Triple P[117] and its modified subsidiary for families where a child has a disability and behavioural problems, Stepping Stones Triple P,[127, 130] were assessed in three studies, with 93 parents randomised to these intervention conditions. The study by Whittingham and colleagues[130] was the only one to include more than one intervention condition, where Stepping Stones Triple P with and without additional ACT both featured. Group play therapy featured in three studies[132, 134, 137] with 44 participants randomised to these treatment arms. Palliative care (n = 49 [122, 136]), Emotional intelligence training (n = 40 [131, 133]) and stress management programmes (n = 16 [119, 121]) featured in two studies each.

Progressive muscle relaxation (PMR) techniques featured in three studies. However, it only formed part of two intervention arms, with 47 participants receiving such interventions. Specifically, Bignall and colleagues[118] trialled an intervention containing breathing retraining with PMR, asthma education and guided imagery, while Yang and colleagues[138] used 30

minutes of audio-taped guided muscle relaxation nightly before going to sleep. PMR was also an active comparator in the intervention trialled by Diego and colleagues[120] where the intervention was massage therapy. Sometimes the intervention label used in the study indicated more than one intervention category might be relevant, for instance Hains and colleagues'[121] cognitive restructuring and problem solving and Nekah and colleagues'[132] structured cognitive-behavioural group play therapy both suggest CBT. We used the description of interventions provided in articles to assess which category was most suitable.

The most common comparator involved treatment as usual (TAU) or usual care (e.g. regular schooling, usual sedation, usual hospital care) within a given setting (n = 20:[82, 83, 117, 119, 121-123, 126, 127, 129-138, 145]), with five of these studies employing a waitlist design.[121, 123, 126, 130, 136] In total, 393 participants were randomised to standard or usual care control groups. Active comparators in the remaining six studies (n = 157 participants) included asthma education,[118] PMR,[120] aerobic exercise[128] and non-directive supportive therapy.[124, 125] Standard care/TAU was often described in only basic detail and varied considerably between studies, to the extent that we were unable to determine common components. During the waiting period of waitlist designs, all participants received usual care.

Interventions were received by parents alone in three studies,[117, 127, 130] parents and children together in five studies,[83, 122, 124, 125, 145] or CYP alone in the remainder. Where both parents and CYP received the intervention, the goal was usually to give parents skills to help them encourage positive behaviours at home, and develop skills to maintain positive effects beyond the intervention end. This occurred as a component of CBT interventions, with the exception of Lyon and colleagues' palliative care intervention,[140] where the aim was to include the family in decision making. The delivery of interventions occurred across a variety of settings and through a range of personnel. Twelve studies delivered at least part of their intervention in a hospital setting.[83, 117, 119, 121, 122, 125, 127-129, 132, 135, 145] Five interventions took place in other medical centres or University outpatient departments,[82, 123, 124, 126, 134] three were delivered in school settings[118, 131, 133] and one at home.[138] In Shoshani and colleagues[136], which assessed the Make a Wish intervention, children's Wishes were ascertained in their own home, but the nature of their delivery was not reported. Seven studies included some component that required either delivery, receipt or practice of the intervention at home.[83, 118, 122, 125-127, 130]

Interventions were delivered by researchers, clinicians, therapists, psychologists, post-graduate students and teachers. Specific training in the delivery of the intervention was reported

in 12 studies.[83, 122, 124-127, 130, 131, 135, 136, 138, 145] Intervention manuals were only referred to in nine studies.[83, 117, 122, 124, 125, 127, 129, 130,126]

Quality appraisal and risk of bias

Table 1 provides a summary of the quality and risk of bias appraisal of included articles. Risk of bias was performed at the article, rather than study level, as data collection and/or outcomes differed across articles and therefore risk of bias may vary where there were several publications reporting on a single study. Eight of the 15 criteria allowed a rating of 'Yes', 'No' or 'unclear'. An additional 'not applicable' option was available for four criteria (whether intention-to-treat analysis was performed, whether the longest follow-up was at six months or longer, whether drop-outs were described, whether missing data were explained). For comparative purposes response rate at longest follow up of 85%+ was considered high, 70-84% was a moderate and <70% was considered a poor response rate. For the assessment of outcome measures, only where all outcomes had good psychometric properties was the score deemed positive.

The included articles were often free from risk of bias on freedom from selective outcome reporting, with 33 of 35 papers rated 'yes'. Lack of differences, or adjustment for differences between groups at baseline was present in 28 of 35 articles. Twenty-seven articles included only outcomes with good psychometric properties, and five of the eight articles not meeting this criterion were marked down on only one outcome measure.[82, 119, 122, 123, 135] Seventeen articles reported a response rate at longest follow-up of 85% or greater; however, none of these articles collected follow-up data at a period six months or later following the intervention. For those eight articles that did include longer follow-ups, response rates were between 70-84% in three articles[117, 142, 145] and <70% in the rest.[127, 129, 130, 139, 144]

The criteria which indicated risk of bias most often across articles were the assessment of adherence, compliance or fidelity (20 of 31 'No'); explaining missing data (13 of 31 'No'); blinding of assessors and inclusion of a follow-up after post-treatment (both 17 of 31 'No'). Additionally, only half of those articles including a follow-up assessment did this at six months or longer. The selection bias domains scored poorly, largely due to lack of clarity in reporting.

Four articles appeared to be of particularly high risk of bias (were judged or poor quality in relation to 50% or more criteria).[82, 126, 134, 135] There were five other articles where fewer than 50% of criteria scored positively, and greater than 15% of criteria were unclear, indicating potential risk of bias.[119, 120, 131, 132, 138] In all five articles, it was not possible to determine whether randomisation was adequate, or allocation was concealed. However, these articles were free from bias in relation to suggestion of selective reporting, and having 85%+ response

rate at follow-up (except Nekah[132]), although none included a follow-up assessment at six months or later.

Both articles by Yetwin[126] and Hains and colleagues[121] were at risk of bias on adequacy of randomisation because both allowed two participants to swap groups after allocation.

The articles demonstrating the lowest risk of bias were by Whittingham and colleagues[130, 144] both of which were rated positively on 13 criteria. The only risk of bias in the Whittingham articles arose because outcomes were self-reported, therefore assessors were not blinded, and the response rate at longest follow up was less than 70 per cent. This poor response rate was symptomatic of the few studies to collect data at the six month follow-up interval. The two articles reporting on the study by Brown and colleagues[127, 139] performed similarly to the Whittingham articles. The article by Reigada and colleagues [124] also showed low risk of bias overall, with only two negative scores.

Table 1: Quality appraisal and risk of bias of included quantitative articles.

	Adequate method of randomisation?	Allocation concealed?	Intention to treat analysis performed?	Blinding of outcome assessor in at least one measure?	Group outcomes similar at baseline, or imbalances accounted for?	Response rate at longest follow up	Intervention well described?	Adherence, compliance or fidelity measured?	Included follow-up beyond post-treatment?	Longest follow-up six months or longer?	Were any drop outs and attrition described	Proportion of outcome measures with good psychometric properties?	Was missing data explained?	Is the study free of suggestion of selective outcome reporting?	Was there a treatment manual for the intervention?
Ashori et al., 2013[146]	?	?	n/a	N	Y	85%+	N	N	N	n/a	n/a	1/1	n/a	Y	N
Bignall et al., 2015[133]	Y	?	N	N	N	85%+	Y	N	N	n/a	Y	4/4	n/a	Y	N
Boardway et al., 1993[134]	?	?	N	?	Y	85%+	Y	N	Y	N	N	11/12	N	Y	N
Brown et al., 2014[142]	Y	Y	Y	N	N	<70%	Y	Y	Y	Y	Y	3/3	Y	Y	Y
Brown et al., 2015[154]	Y	Y	Y	N	N	<70%	Y	Y	Y	Y	Y	7/7	Y	Y	Y
Bufalini, 2009[150]	?	?	n/a	N	N	85%+	N	N	N	n/a	n/a	3/4	n/a	Y	N
Diego et al., 2001[135]	?	?	n/a	?	?	85%+	Y	N	N	n/a	n/a	4/4	n/a	Y	N
Gordon et al., 2010[143]	Y	Y	Y	Y	N	85%+	N	N	N	n/a	Y	5/7	Y	Y	N
Hains et al., 2000[136]	N	?	N	?	Y	85%+	Y	N	Y	N	n/a	5/5	N	Y	N
Lyon et al., 2014[137]	Y	?	Y	N	Y	85%+	Y	N	N	n/a	n/a	4/4	Y	Y	Y
Lyon et al., 2013[155]	Y	?	Y	N	Y	85%+	Y	Y	N	n/a	n/a	2/3	Y	Y	Y

	Adequate method of randomisation?	Allocation concealed?	Intention to treat analysis performed?	Blinding of outcome assessor in at least one measure?	Group outcomes similar at baseline, or imbalances accounted for?	Response rate at longest follow up	Intervention well described?	Adherence, compliance or fidelity measured?	Included follow-up beyond post-treatment?	Longest follow-up six months or longer?	Were any drop outs and attrition described	Proportion of outcome measures with good psychometric properties?	Was missing data explained?	Is the study free of suggestion of selective outcome reporting?	Was there a treatment manual for the intervention?
Martinović et al., 2006[97]	Y	?	n/a	N	Y	85%+	N	N	Y	N	N	4/5	n/a	Y	N
Masia Warner et al., 2011[138]	Y	?	N	Y	Y	85%+	N	N	Y	N	Y	4/5	Y	Y	N
Nekah et al., 2015[147]	?	?	N	N	Y	70-84%	Y	N	N	N	Y	1/1	n/a	Y	N
Pourmohamadreza-Tajrishi et al., 2013[148]	?	?	n/a	N	Y	85%+	N	N	N	n/a	n/a	1/1	n/a	Y	N
Reigada et al., 2015[139]	Y	?	Y	Y	Y	85%+	Y	Y	Y	N	Y	4/4	N	Y	Y
Serlachius et al., 2014[144]	Y	Y	Y	?	Y	<70%	Y	N	Y	Y	N	4/4	N	Y	Y
Shoshani et al., 2015[151]	Y	?	N	N	Y	85%+	N	N	Y	N	Y	5/5	N	Y	N
Szigethy et al., 2007[98]	?	?	N	Y	Y	<70%	Y	Y	N	n/a	Y	6/6	N	Y	Y

	Adequate method of randomisation?	Allocation concealed?	Intention to treat analysis performed?	Blinding of outcome assessor in at least one measure?	Group outcomes similar at baseline, or imbalances accounted for?	Response rate at longest follow up	Intervention well described?	Adherence, compliance or fidelity measured?	Included follow-up beyond post-treatment?	Longest follow-up six months or longer?	Were any drop outs and attrition described	Proportion of outcome measures with good psychometric properties?	Was missing data explained?	Is the study free of suggestion of selective outcome reporting?	Was there a treatment manual for the intervention?
Szigethy et al., 2009[156]	?	?	N	Y	Y	<70%	Y	Y	N	n/a	N	3/3	N	Y	Y
Szigethy et al., 2014[140]	?	?	Y	Y	Y	70-84%	Y	Y	N	n/a	N	7/7	N	Y	Y
Szigethy et al., 2015[158]	?	?	Y	Y	Y	70-84%	Y	Y	N	n/a	N	5/5	N	N	Y
Thompson et al., 2012[157]	?	?	N	Y	Y	70-84%	Y	Y	Y	Y	N	3/3	N	Y	Y
Wang et al., 2012[152]	Y	?	N	N	Y	85%+	Y	N	N	n/a	Y	2/2	n/a	Y	N
Westrupp et al., 2015[132]	Y	Y	Y	N	Y	70-84%	N	N	Y	Y	N	9/9	N	Y	Y
Whittingham et al., 2014[145]	Y	Y	Y	N	Y	<70%	Y	Y	Y	Y	Y	3/3	Y	Y	Y
Whittingham et al., 2015[159]	Y	Y	Y	N	Y	<70%	Y	Y	Y	Y	Y	4/4	Y	Y	Y
Wicksell et al., 2009[160]	Y	?	Y	N	Y	70-84%	Y	N	Y	Y	N	7/9	N	Y	N

	Adequate method of randomisation?	Allocation concealed?	Intention to treat analysis performed?	Blinding of outcome assessor in at least one measure?	Group outcomes similar at baseline, or imbalances accounted for?	Response rate at longest follow up	Intervention well described?	Adherence, compliance or fidelity measured?	Included follow-up beyond post-treatment?	Longest follow-up six months or longer?	Were any drop outs and attrition described	Proportion of outcome measures with good psychometric properties?	Was missing data explained?	Is the study free of suggestion of selective outcome reporting?	Was there a treatment manual for the intervention?
Yang et al., 2004[153]	?	?	n/a	Y	Y	85%+	N	N	N	n/a	n/a	2/8	n/a	Y	N
Yetwin 2011[141]	N	?	N	?	Y	70-84%	N	N	N	n/a	Y	8/8	N	N	Y
Zareapour, 2009[149]	?	?	N	N	Y	85%+	N	N	N	N	Y	1/1	n/a	Y	N

Y = yes; N = no; ? = unclear; n/a = not applicable; green cell denotes positive score; yellow cell denotes unclear or moderate score; red cell denotes negative score.

Analysis of included study findings

Effectiveness of cognitive behavioural therapy interventions

Interventions categorised as CBT were the most common type of intervention, featuring in ten articles reporting on seven studies across a range of LTCs: epilepsy; persistent functional somatic complaints; IBD; T1DM and chronic pain.[82, 83, 123-125, 129, 145] Across the seven studies, 531 participants were randomised. Comparators were either TAU, waitlist or non-directive supportive therapy (NDST).[83, 124, 125] Despite being the most common intervention type, there was no opportunity to meta-analyse data due to differing LTCs, control groups and outcomes. The included studies were also characterised by small samples and varying risk of bias. Six different CBT interventions were assessed across the seven studies. All interventions in this category shared components of typical CBT interventions and/or were identified as a CBT intervention. All these interventions aimed to improve both mental and physical health. Mental health variables included depression,[82, 83, 125, 145] anxiety[123, 124] and stress.[129] Parent sessions were included in all but two of the interventions.[82, 129] Exposure, and ACT in Wicksell and colleagues'[145] study was categorised as CBT because exposure exercises were seen in other CBT interventions,[123, 124] and the authors[145] characterise ACT as a development of CBT.

Five of the interventions (Best of Coping,[129] TAPS,[123] TAPS+IBD,[124] PASCET-PI,[83, 125] Exposure and ACT[145]) contained content adapted for the LTC of the sample in the study (T1DM,[129] Persistent functional somatic complaints,[123] IBD[83, 124, 125] or chronic pain[145]) by the inclusion of intervention content such as: tasks identifying IBD-specific stressors and developing ways to cope with symptoms,[124] addressing fears specifically related to physical pain[123] and integrating illness narratives and working on the development of healthy IBD-related cognitions and behaviours. The intervention trialled by Martinović and colleagues targeted depressive thoughts, and did not contain any LTC-specific (epilepsy) content.[82]

Children and young people mental health outcomes

Depression was assessed in the four studies whose intervention aimed to improve this outcome.[82, 83, 125, 145] Three of the studies measured depression using multiple measures,[82, 83, 125] meaning this was the most frequently occurring outcome category for CBT interventions. General mental health was assessed in four studies,[83, 123, 125, 145] and two studies whose interventions aimed to improve anxiety also measured it as an outcome.[123, 124]

Supplementary Materials 1, 6 displays means and SD for control and intervention groups for each CYP mental health outcome assessed at post-intervention, where sample size, mean and SD were reported or calculable. Effect sizes (Cohen's *d*), 95% confidence intervals and *p*-values are included, with a positive effect size representing improvement on the measure.

Evidence of the beneficial effect of CBT for measures of CYP mental health can be found in six of the seven studies featuring this type of intervention. Evidence of the beneficial effect of CBT on depression across all outcomes is provided by one study,[82] where large positive effect sizes are seen for all depression measures, although wide 95% confidence intervals for two of the measures include negligible effect sizes reflecting the imprecision of the estimate of effect (Beck Depression Inventory: d = 0.85; 95% CI: 0.10 to 1.60, p = 0.03; Centre for Epidemiological Studies Depression scale: d = 0.86; 95% CI: 0.11 to 1.61, p = 0.03). Evidence for the beneficial effect of CBT on depression from other studies is less clear; while small to large positive effect sizes were seen for the other studies reporting depression outcomes, these effects tended to be imprecise with 95% confidence intervals typically including negligible or even slightly harmful effects. Reigada[124] provides evidence of a large beneficial effect of CBT on IBD-specific anxiety (d = 1.31; 95% CI: 0.36 to 2.27, p = 0.007). However, there was little evidence for a beneficial effect of CBT on anxiety in the study by Masia Warner and colleagues[123] (d = 0.27; 95% CI: -0.38 to 0.92, p = 0.41). There was some evidence for a large beneficial effect of CBT on general mental health according to Masia Warner and colleagues[123] (d = 1.11; 95% CI: 0.42 to 1.81, p = 0.002) and Szigethy and colleagues[83] (d = 0.91; 95% CI: 0.25 to 1.56, p = 0.007), although the wide confidence intervals in the latter include small effect sizes reflecting the imprecision of the estimate of effect. However, Wicksell and colleagues[145] report a lack of evidence for the beneficial effect of CBT on general mental health (d = 0.40; 95% CI: -0.40 to 1.13, p = 0.41) in their trial. There was no opportunity to meta-analyse CYP mental health outcomes for these interventions evaluating CBT due to differing LTCs or comparators across studies where outcome categories were shared.

Other outcomes

Supplementary Materials 1, displays findings for other outcomes assessed at post-intervention. Conflicting evidence regarding the beneficial effect of CBT on LTC-specific quality of life and LTC symptoms can be seen across the studies featuring CBT interventions. For instance, while the study by Martinović and colleagues[82] provides evidence of a large beneficial effect of CBT for epilepsy-specific quality of life (d = 1.55; 95% CI: 0.72 to 2.37, p < 0.001), there was a lack of evidence for such a beneficial effect on diabetes control reported by Serlachius and colleagues[129] (d = 0.32; 95% CI: -0.08 to 0.73, p = 0.12). The study by Masia Warner and colleagues[123] provides evidence of some medium to large beneficial effect sizes for CBT on measures of LTC symptoms. Large beneficial effect sizes for self-reported (d = 1.33; 95% CI: 0.61 to 2.05, p < 0.001) and parent-reported pain (d = 0.98; 95% CI: 0.30 to 1.67, p = 0.005) were found, as well as a medium beneficial effect on somatisation (d = 0.75; 95% CI: 0.08 to 1.42, p = 0.03), although the wide confidence interval for somatisation includes negligible effect sizes reflecting the imprecision of this estimate of effect. The study by Wicksell and colleagues[145] provides evidence of large beneficial effects of CBT for pain (d = 1.25; 95% CI: 0.45 to 2.05, p = 0.002) and pain-related emotional discomfort (d = 1.46; 95% CI: 0.63 to 2.28, p < 0.001). However, there was little evidence for the beneficial effect of CBT on other pain and LTC symptoms outcomes measured by Wicksell and colleagues.[145] There was little evidence for the beneficial effect of CBT on LTC symptoms in other studies.[124, 125, 129]

Evidence for the effect of CBT on LTC symptom severity could be investigated further by meta-analysing outcomes from Szigethy and colleagues and Reigada and colleagues,[124, 125] where both samples had IBD and the comparator was NDST. Reigada and colleagues reported pooled data for the PUCAI and PCDAI (Paediatric Ulcerative Colitis/Crohn's Disease Activity Index respectively) outcomes within the paper,[124] whereas Szigethy and colleagues[125] reported separately for participants with ulcerative colitis and Crohn's disease respectively.

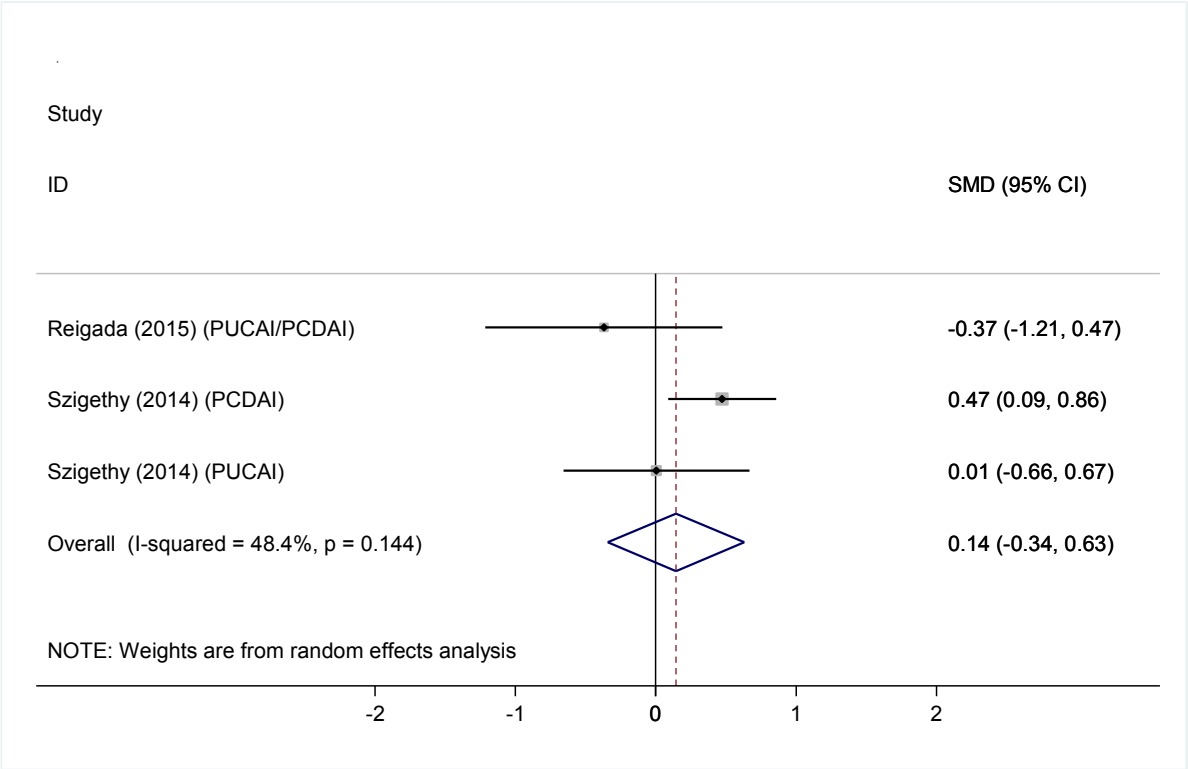

Figure 3 shows a forest plot for the effects of CBT on these disease activity measures for these studies. This meta-analysis demonstrated little evidence of an effect of CBT on LTC symptoms (d = 0.14; 95% CI: -0.34 to 0.63, p = 0.56).

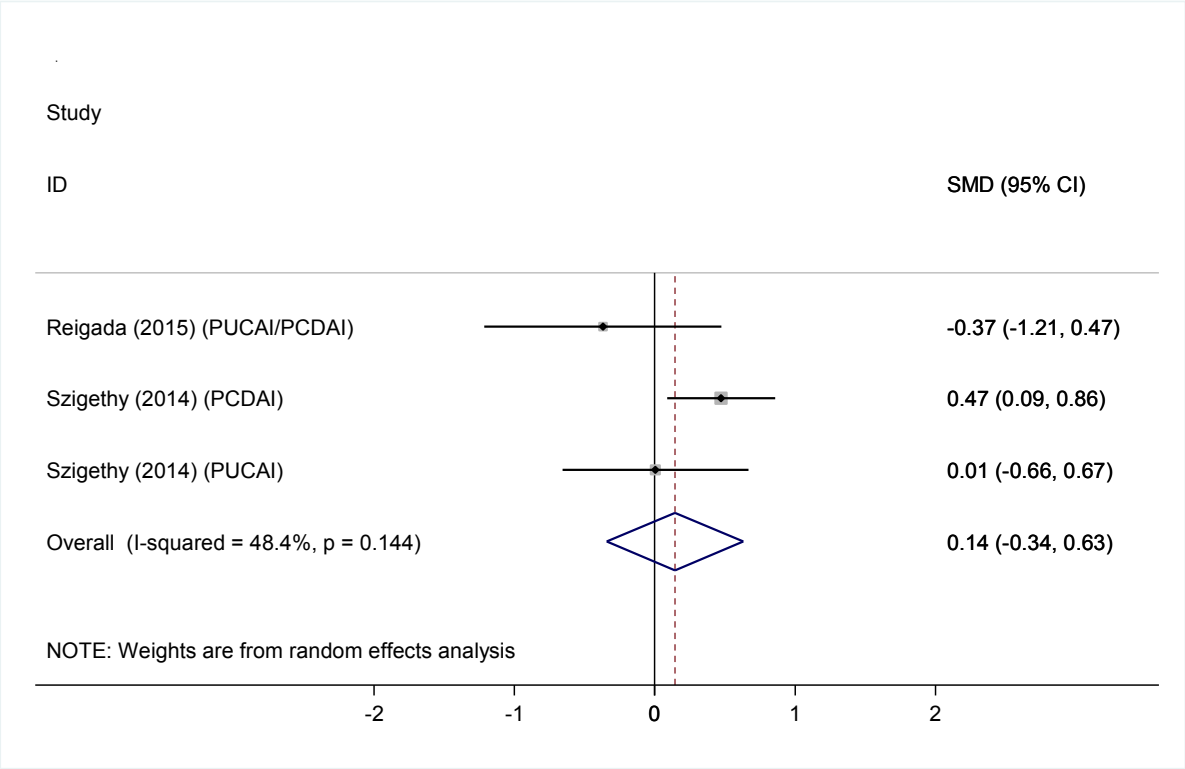

Figure 3: Forest plot showing the results of meta-analysis of the effects of CBT on IBD symptoms at post-intervention for included studies. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; PUCAI = Pediatric Ulcerative Colitis Activity Index; PCDAI = Pediatric Crohn's Disease Activity Index.

Overall, the seven included studies that evaluated the effectiveness of interventions categorised as CBT aiming to improve the mental health of CYP with LTCs show some promising effects. A number of large beneficial effects for CYP mental health and other outcomes were seen, although effects for particular outcomes were rarely consistent across all studies. The heterogeneity in study characteristics prevented meta-analysis of CYP mental health outcomes. The available sample of studies is small, targeting different aspects of mental health, for children with different LTCs. There was also diversity in quality and risk of bias across these studies.

Effectiveness of parenting interventions

Parenting interventions without acceptance and commitment therapy

Of the four parenting interventions assessed in included studies, two were parenting without ACT which were assessed by Westrupp and colleagues[117] and Whittingham and colleagues.[130, 144] Westrupp and colleagues assessed the *Positive Parenting Programme* (Triple

P) in families with a diabetic child, while one of the intervention arms in Whittingham and colleagues was the 'Stepping Stones' version of Triple P (SSTP) alone. SSTP is tailored towards families with pre-adolescent children with a disability, which may be physical or intellectual, and behavioural problems.[147] The sample in the study by Whittingham and colleagues had Cerebral Palsy.[130] Both studies had similar aims including improving aspects of CYP mental health, with a focus on behavioural and emotional problems in Whittingham and colleagues, as well as improving parenting. A total of 50 participants were randomised to intervention or control arms, with all control participants (n = 32) receiving TAU (Whittingham and colleagues used a waitlist control). In both studies, the primary aim was to reduce child behavioural problems through improved parenting skills. Westrupp and colleagues examined the effects of Triple P in a group of families with children who had elevated behavioural problems at baseline, as well as a group without elevated behavioural symptoms.[117] We were unable to obtain raw data for the sub-group analysis of families with children with behaviour problems, and therefore could not calculate effect sizes for the study.

Children and young people ***mental health outcomes***

Supplementary Materials 1, 8 displays findings for each CYP mental health outcome assessed at post-intervention by Whittingham and colleagues.[130] Despite a medium sized beneficial effect for the ECBI: Problems subscale (d = 0.72; 95% CI: 0.04 to 1.40, p = 0.04), there was a lack of evidence for the beneficial effect of a parenting programme on child behaviour, with 95% confidence intervals on the other measures of child behaviour all including harmful effects. There was conflicting evidence of the effect of parenting on social dysfunction, where a large beneficial effect was seen for the SDQ peer problems subscale, although wide 95% confidence interval includes negligible effect sizes reflecting the imprecision of the estimate of effect (d = 0.88; 95% CI: 0.19 to 1.57, p = 0.01), but there was little evidence for its beneficial effect on the prosocial subscale (d = -0.19; 95% CI: -0.84 to 0.47]. A medium effect size was also found in this study for SDQ emotional behaviour subscale (d = 0.64; 95% CI: -0.03 to 1.31, p = 0.06) but the imprecise 95% confidence interval includes negligibly harmful effects. While it was not possible to calculate effect sizes for the outcomes reported by Westrupp and colleagues,[117] the article reports medium to large and statistically significant beneficial effects for externalising problems, internalising problems and disruptive behaviours for the participants with behaviour problems receiving the intervention. Because of the different LTCs in the two studies and the lack of necessary statistics reported in the study

by Westrupp and colleagues,[117] meta-analysis was not performed for these CYP mental health outcomes or others reported below.

Other outcomes

Numerous secondary outcomes were also reported. Given the parent-focused nature of the interventions, the studies by both Westrupp and colleagues and Whittingham and colleagues assessed parenting styles and parent mental health.[117, 144] A number of outcomes related to LTC symptoms and LTC-specific quality of life were also reported. *Supplementary Materials 1, 9* displays findings for these other outcomes. Overall the study by Whittingham and colleagues demonstrates a lack of evidence of a beneficial effect of this parenting intervention on other outcomes, except for parenting confidence, which recorded a medium sized beneficial effect but the wide 95% confidence interval included negligible effect sizes (d = 0.69; 95% CI: 0.02 to 1.37, $p < 0.05$). [130, 144] . In the article by Westrupp and colleagues,[117] findings showed statistically significant medium effect sizes for parent mental health but only evidence of the benefit of a parenting intervention for some measures of parenting and family functioning for participants with behaviour problems.

Follow up data

Westrupp and colleagues[117] included follow up assessments of all outcomes at 12 months post-intervention. Their article reports that large statistically significant favourable effects were seen for parent anxiety and stress at 12 months, but not for the CYP mental health and other outcomes assessed.

These two studies investigated the effect of parenting programmes on a wide range of outcomes, yet clear evidence for benefits was rarely seen. The study by Whittingham and colleagues provides conflicting evidence relating to child behaviour and social dysfunction across different measures used.[130] For other outcomes, reported effect sizes were typically small with wide confidence intervals reflecting imprecision in effect estimates. Overall, with only two RCTs assessing parenting interventions without an ACT component and only one providing data that allowed the calculation of effect sizes, there is weak evidence relating to the effectiveness of parenting programmes for improvement of child or parent outcomes.

Parenting interventions with acceptance and commitment therapy

The study by Whittingham and colleagues included two intervention groups; as well as SSTP summarised above, the authors assessed the effectiveness of SSTP plus ACT.[130] Brown and colleagues also evaluated the effectiveness of SSTP+ACT.[127] Whittingham and colleagues investigated SSTP+ACT in children with Cerebral Palsy, while the sample of children in the trial by Brown and colleagues had an acquired brain injury, of whom a proportion (seven per cent) had Cerebral Palsy. Both studies compared the parenting intervention to TAU, with Whittingham and colleagues employing a waitlist control group.[130] Both studies aimed to reduce child behaviour and emotional problems and dysfunctional parenting. Because of the consistent comparator and LTC between studies, similar outcomes were considered eligible for meta-analysis. In total, the parents of 92 children were randomised evenly between SSTP+ACT and control groups. Whittingham was a co-author on both Brown et al. papers while Boyd and McKinlay were co-authors on all four included papers across both studies.[127, 130, 139, 144]

Children and young people ***mental health outcomes***

Supplementary Materials 1, Table 10 summarises findings for each CYP mental health outcome assessed at post-intervention. As for the SSTP intervention discussed in the previous section, evidence of improvement on the ECBI: Problems subscale is provided in the study by Whittingham and colleagues[130] where a large beneficial effect size was observed (d = 1.34; 95% CI: 0.65 to 2.03, p < 0.001). There was a lack of evidence for the beneficial effect of SSTP+ACT on other behaviour outcomes. Only for the ECBI: Intensity subscale was a medium beneficial effect reported, but the wide 95% confidence interval included negligible beneficial effect sizes (d = 0.78; 95% CI: 0.14 to 1.43, p = 0.02). Brown and colleagues observed a slightly larger effect for the same measure (d = 0.82; 95% CI: 0.25 to 1.38, p = 0.005),[127] although they did not mirror the evidence from Whittingham and colleagues'[130] study of a large

beneficial effect for the ECBI: Problems scale. Brown and colleagues also found a medium beneficial effect for a measure of emotion problems (d = 0.76; 95% CI: 0.19 to 1.32, p = 0.009), although the wide confidence interval reflects the imprecision of the estimate. There was little evidence for a similar beneficial effect for emotional problems in the study by Whittingham and colleagues.[130]

As these studies had the same intervention, LTC and comparator, meta-analysis was performed on behaviour (see *Figure 4*), and emotional problems outcomes (see *Figure 5*). For the meta-analysis of child behaviour the effects from ECBI: Problems and ECBI: Intensity were combined into a single summary effect for each study, calculating the standard error for this effect using an estimate of the correlation between the subscales, obtained from other research.[148] There was evidence of the beneficial effect of SSTP+ACT for both outcomes with a large beneficial effect for child behaviour problems (d = 0.81; 95% CI: 0.38 to 1.23, p < 0.001) and a medium beneficial effect for emotional problems, although the latter in particular was characterised by wide confidence intervals that included small effect sizes (d = 0.58; 95% CI: 0.17 to 1.00, p < 0.006). As both Brown and colleagues[127] and Whittingham and colleagues[130] used the SDQ: Emotional problems subscale, meta-analysis using mean differences can be seen in *Appendix 6*.

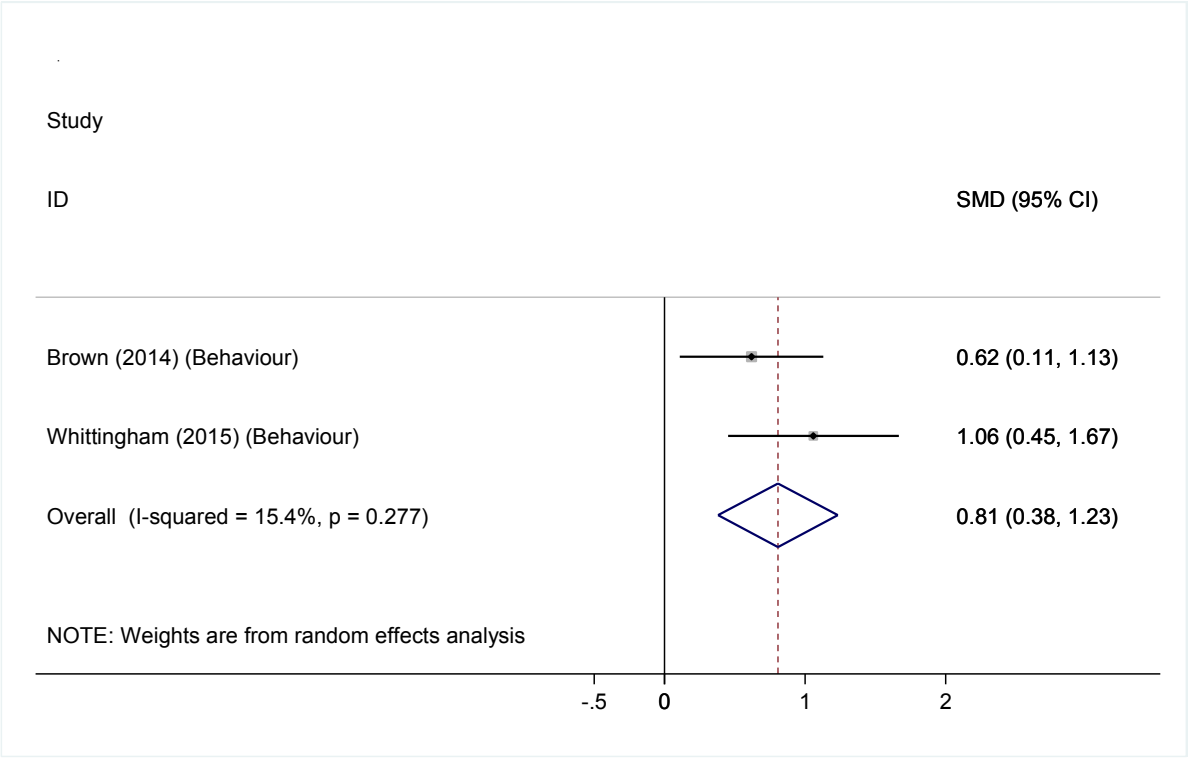

Figure 4: Forest plot showing the results of meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on child behaviour at post-intervention for CYP with acquired brain injury and/or Cerebral Palsy. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval.

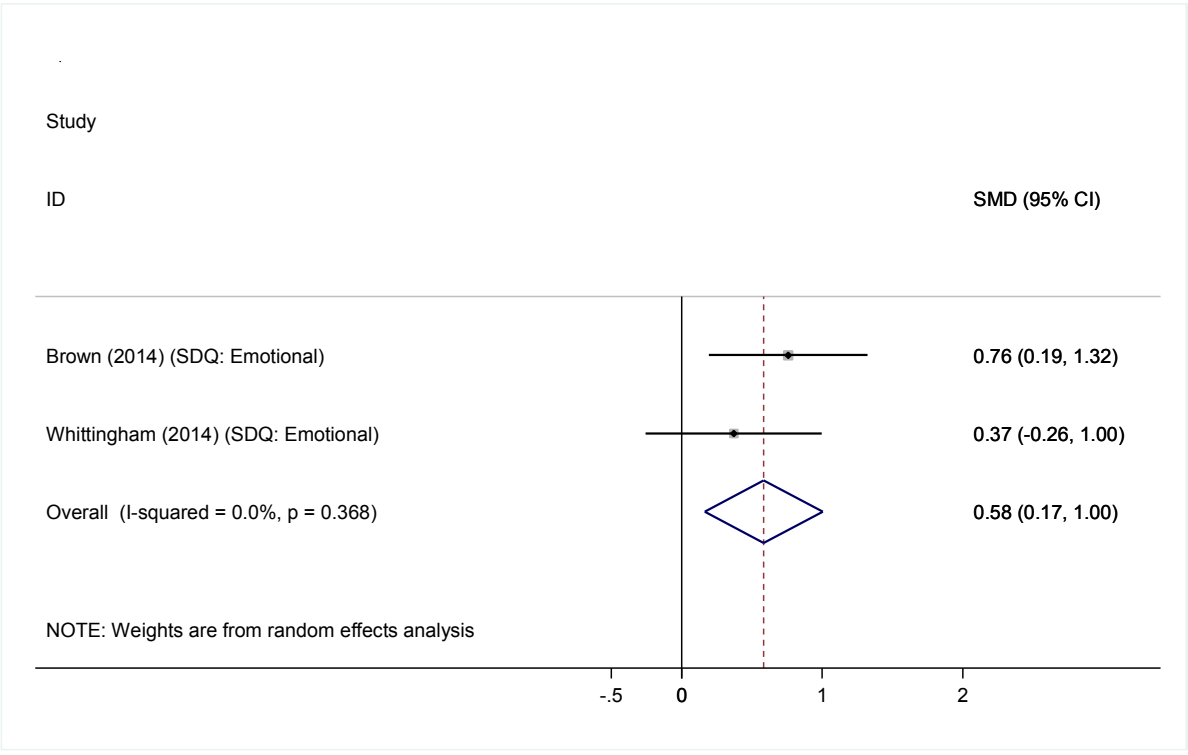

Figure 5: Forest plot showing the results of meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on child emotional problems at post-intervention for CYP with acquired brain injury and/or

Cerebral Palsy. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; SDQ = Strengths and Difficulties Questionnaire.

Other outcomes

Supplementary Materials 1, displays findings for other outcomes assessed at post-intervention in these studies. There is some evidence for the beneficial effect of SSTP+ACT for some outcomes relating to parenting styles, parenting health and LTC-specific quality of life. The largest effect size was for parenting laxness (a measure of parents' inconsistent or permissive parenting style) (d = 1.20; 95% CI: 0.61 to 1.79, $p < 0.001$) in the study by Brown and colleagues,[127] although this was not mirrored in Whittingham and colleagues' study.[130] Parenting over-reactivity was associated with a large beneficial effect in both Brown and colleagues (d = 0.83, 95% CI: 0.26 to 1.40, p = 0.004) and Whittingham and colleagues studies (d = 1.11, 95% CI: 0.44 to 1.78, p = 0.001).[130, 144] There was further evidence for the beneficial effect of SSTP+ACT on parenting provided by Brown and colleagues (Parent Thoughts and Feelings Questionnaire: d = 0.88, 95% CI: 0.31 to 1.45; Parenting Tasks Checklist Behaviour subscale: d = 0.91, 95% CI: 0.33 to 1.48).[139] A range of other measures of parenting were used in both studies and many others provided medium to large effects, although these effects tended to be imprecise with 95% confidence intervals typically including negligible effect sizes. There was less evidence from Whittingham and colleagues[130] for the beneficial effect of SSTP+ACT on LTC-specific QoL and LTC symptoms.

Meta-analysis was performed for parent mental health (see *Figure 6*) and parenting outcomes (see *Figure 7*). The range of individual measures reported above for each outcome category, were combined into a single summary effect for each study. There was evidence for the beneficial effect of SSTP+ACT on parent mental health with a medium sized effect (d = 0.57; 95% CI: 0.30 to 0.83, $p < 0.001$) and a large beneficial effect size was reported for parenting (d = 0.90; 95% CI: 0.57 to 1.24, $p < 0.001$). Meta-analysis using mean differences for parent mental health and parenting outcomes can be seen in *Appendix 6*.

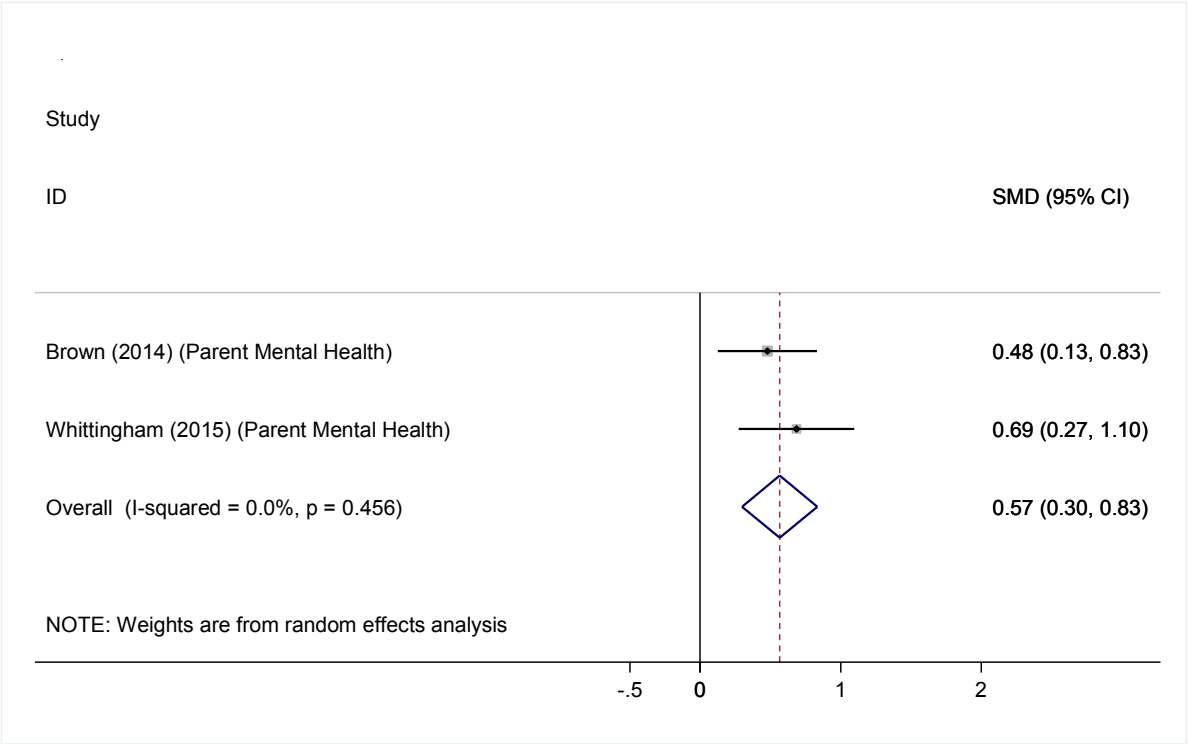

Figure 6: Forest plot showing the results of meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parent mental health at post-intervention for parents of CYP with acquired brain injury and/or Cerebral Palsy. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval.

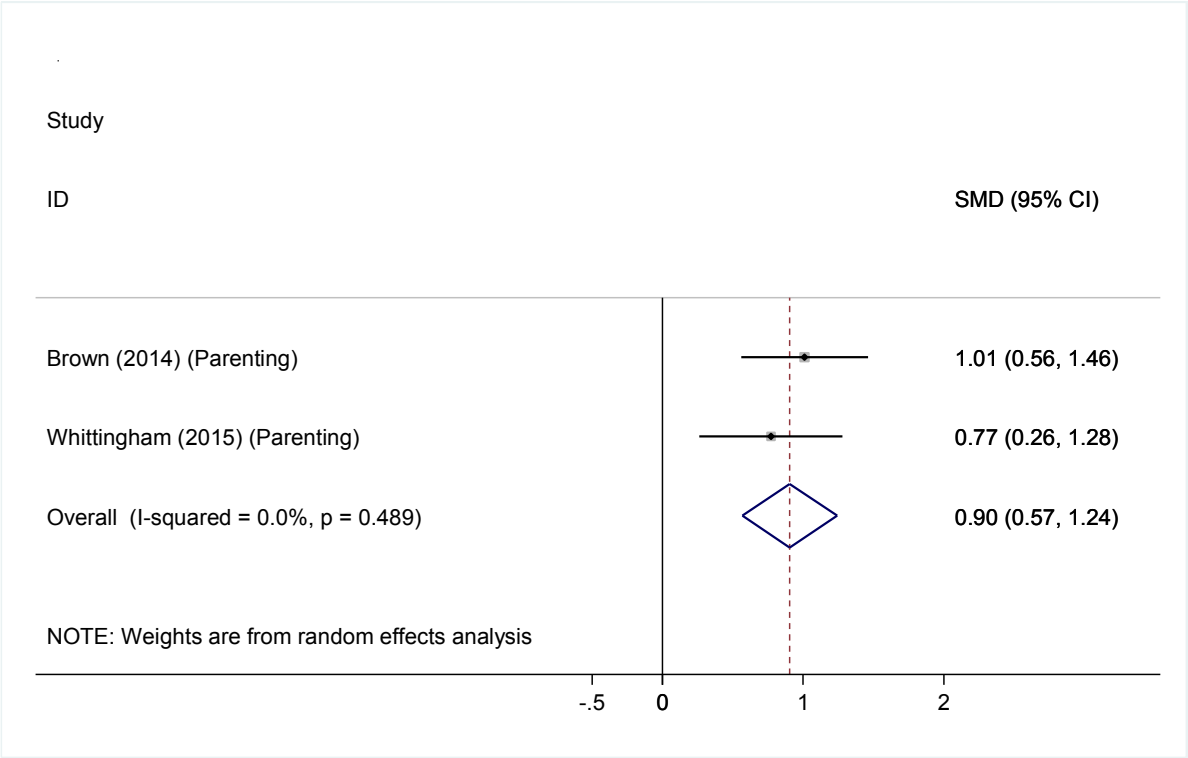

Figure 7: Forest plot showing the results of meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parenting at post-intervention for parents of CYP with acquired brain injury and/or Cerebral Palsy. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval.

The two studies that assessed the effectiveness of SSTP+ACT[127, 130] provide tentative evidence that it may have beneficial effects for some outcomes, including child behaviour, emotional problems, parenting and parent mental health. While the meta-analyses summarise these overall effects, there were contrasting effects from the range of different measures used to quantify the effectiveness of SSTP+ACT, so conclusions relating to more fine-grained outcomes are unclear. Notwithstanding the small number and size of trials of parenting interventions, the findings may indicate the benefit of the addition of ACT to the SSTP programme. Although the included articles scored well on quality appraisal, it is noted that there was some risk of bias because blinding of participants was not possible, and outcomes were rated by parents.[127, 130, 139, 144]

Overall, there is a lack of research investigating the effectiveness of parenting programmes that measured CYP mental health outcomes aside from Triple-P. We know from other systematic reviews that trials have delivered other parenting programmes, but not measured the effects on child mental health.[72]

Effectiveness of group play therapy interventions

Three studies evaluated the effectiveness of group play therapy interventions, with Nekah and colleagues, and Zareapour and colleagues studying children with cancer and elevated symptoms of depression, and Wang and colleagues sampling children with asthma and behavioural problems.[132, 134, 137] All three group play therapy studies took place in non-OECD countries (Iran and China) and were translated from Persian or Chinese. The study by Zareapour and colleagues utilised daily art-based play over the course of a week.[134] Art and crafts featured in Nekah and colleagues' trial of a play programme, which also contained aspects of storytelling and performance, supplemented with a behavioural therapy focus.[132] Wang and colleagues placed greater emphasis on team building, development of confidence and learning through games and activities in their trial.[137] A total of 88 participants were included, of which 48 were randomised to the intervention arms. All control participants received TAU. The study by Zareapour and colleagues aimed to improve depression primarily,[134] while Nekah and colleagues targeted depression and anxiety.[132] Wang and colleagues aimed to improve mental coping ability in their trial.[137] It is notable that the studies reporting group play therapy interventions scored relatively poorly on quality.

Children and young people ***mental health outcomes***

Supplementary Materials 1, Table 12 displays findings for each CYP mental health outcome assessed at post-intervention. Nekah and colleagues observed a large, beneficial effect for anxiety, however the wide confidence interval includes negligible effect sizes and raises uncertainty about the effect (d = 1.29; 95% CI: 0.11 to 2.46, p = 0.04).[132] Similarly this study reported a large beneficial effect for depression, although the wide confidence interval include slightly harmful effects (d = 1.04; 95% CI: -0.10 to 2.17, p = 0.08). On the other hand, the study by Zareapour and colleagues reported a particularly large, beneficial effect on depression as a result of art-based group play therapy (d = 3.40; 95% CI: 2.12 to 4.69, $p < 0.001$).[134] In the study by Wang and colleagues, coping improved with a large effect size (d = 0.93; 95% CI: 0.27 to 1.59, p = 0.006), but there was little evidence for the beneficial effect of the intervention on behaviour problems (d = 0.31; 95% CI: -0.32 to 0.94, p = 0.32).[137]

Given that Nekah and colleagues and Zareapour and colleagues both studied samples with cancer and TAU control groups, meta-analysis was possible for their depression outcomes.[132, 134] *Figure 8* shows a forest plot for the effects of group play therapy on child depression at post-intervention. Meta-analysis revealed that the large beneficial effect size was characterised by very wide confidence intervals that include slightly harmful effect sizes (d = 2.20; 95% CI: -0.12 to 4.52, p = 0.06) and large heterogeneity between the two studies' findings, (I^2 = 86.3%), limiting the evidence for positive effects.

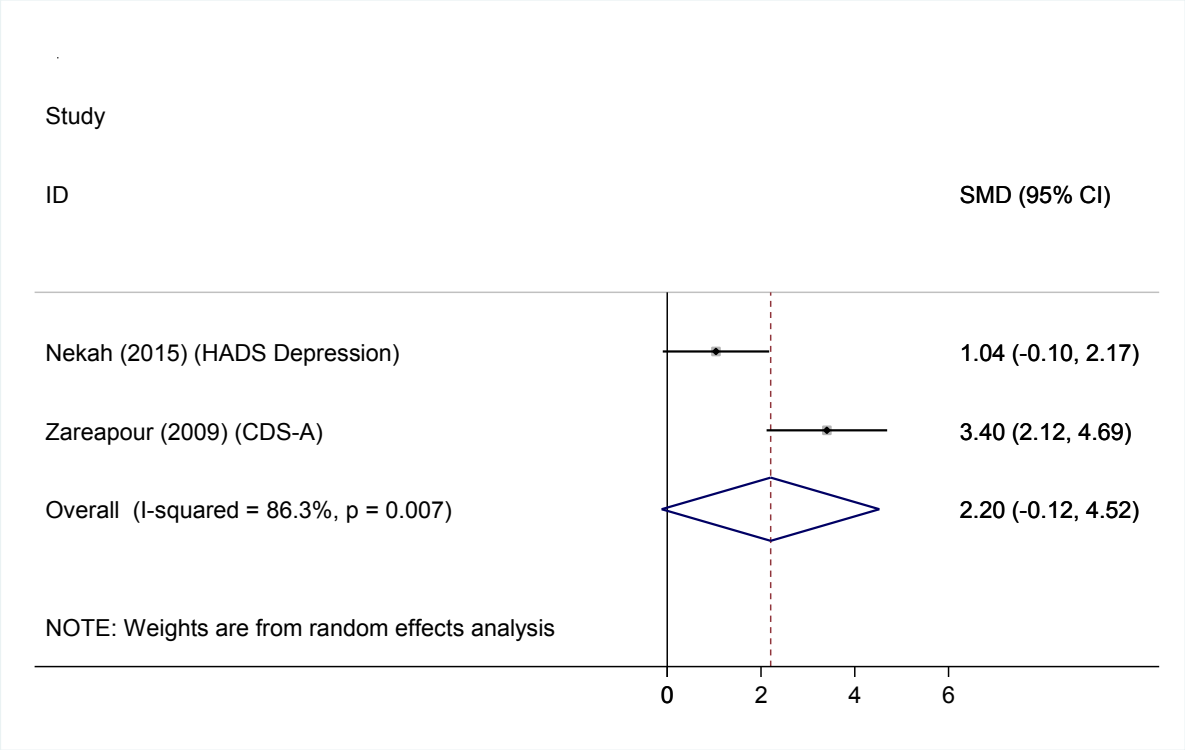

Figure 8: Forest plot showing the results of meta-analysis of the effects of group play therapy on child depression at post-intervention for CYP with cancer. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval. HADS = Hospital Anxiety and Depression Scale; CDS-A = Child Depression Scale-A.

Overall, the included studies cannot tell us with precision whether group play therapy interventions are beneficial for depression or anxiety. Despite the individual findings of a large beneficial effect in the study by Zareapour and colleagues, when meta-analysed, this effect is uncertain.[134] Behaviour outcomes did not improve in the study by Wang and colleagues; however, there may be a beneficial effect for coping.[137] The low quality and high risk of bias across the three studies further weakens the strength of conclusions drawn from the evidence.

Effectiveness of palliative care interventions

Two studies evaluated the effectiveness of interventions which we have categorised broadly as palliative care, both studying a sample of children with cancer.[122, 140, 136] Lyon and colleagues evaluated Family Centred Advance Care Planning for Teens with Cancer (FACE-TC) and elevated symptoms of anxiety,[122] while Shoshani and colleagues evaluated the Make a Wish programme in CYP with mixed mental health symptoms (anxiety, depression, general mental health).[136] FACE-TC placed emphasis on a family-centred approach to discussions about the future, whereas the Make a Wish programme aimed to identify and deliver an activity that the child wanted, without influence from their parents. While these interventions clearly differ in content, they both fit broad definitions of palliative care[149] and share some intervention targets including hope. Control participants in both studies received TAU, with Shoshani and colleagues employing a waitlist design in their study.[136] A total of 96 participants were included, of which 47 were randomised to the intervention arms. The majority of outcomes were assessed three months post-intervention in the study by Lyon and colleagues and five weeks post-Wish in Shoshani and colleagues' study. There were no further follow-up assessments.

Children and young people mental health outcomes

Both palliative care studies assessed child anxiety and depression. Shoshani and colleagues also measured emotional problems, hope and optimism. *Supplementary Materials 1, 13* displays findings for CYP mental health outcomes assessed at post-intervention. Taking the results of Lyon and colleagues' trial in isolation gives little evidence of the beneficial effect of FACE-TC for anxiety and depression outcomes.[122] The trial by Shoshani and colleagues found that the Make a Wish intervention led to a large, beneficial effect for positive emotions (d = 1.03; 95% CI: 0.52 to 1.55, $p < 0.001$), but a lack of evidence for reduction in negative emotions.[136] Shoshani and colleagues also reported a range of medium, beneficial effects for anxiety, depression, general mental health and hope, but all of these effects were characterised by imprecision, with wide confidence intervals including small effect sizes.

Given the similarities in study characteristics, meta-analyses were conducted where outcome categories were shared. *Figure 9* shows a forest plot for the effects of palliative care on child anxiety at post-intervention for Lyon and colleagues and Shoshani and colleagues.[122, 136] *Figure 10* shows a forest plot for the effects of palliative care on child depression at post-intervention for these studies. Meta-analysis revealed that palliative care interventions show a medium-sized, beneficial effect for reducing both child anxiety (d = 0.61; 95% CI: 0.19 to 1.02, p = 0.004) and depression (d = 0.61; 95% CI: 0.19 to 1.03, p = 0.05). However, there is uncertainty in concluding a positive effect of palliative care interventions for these aspects of child mental health as wide confidence intervals include small effect sizes.

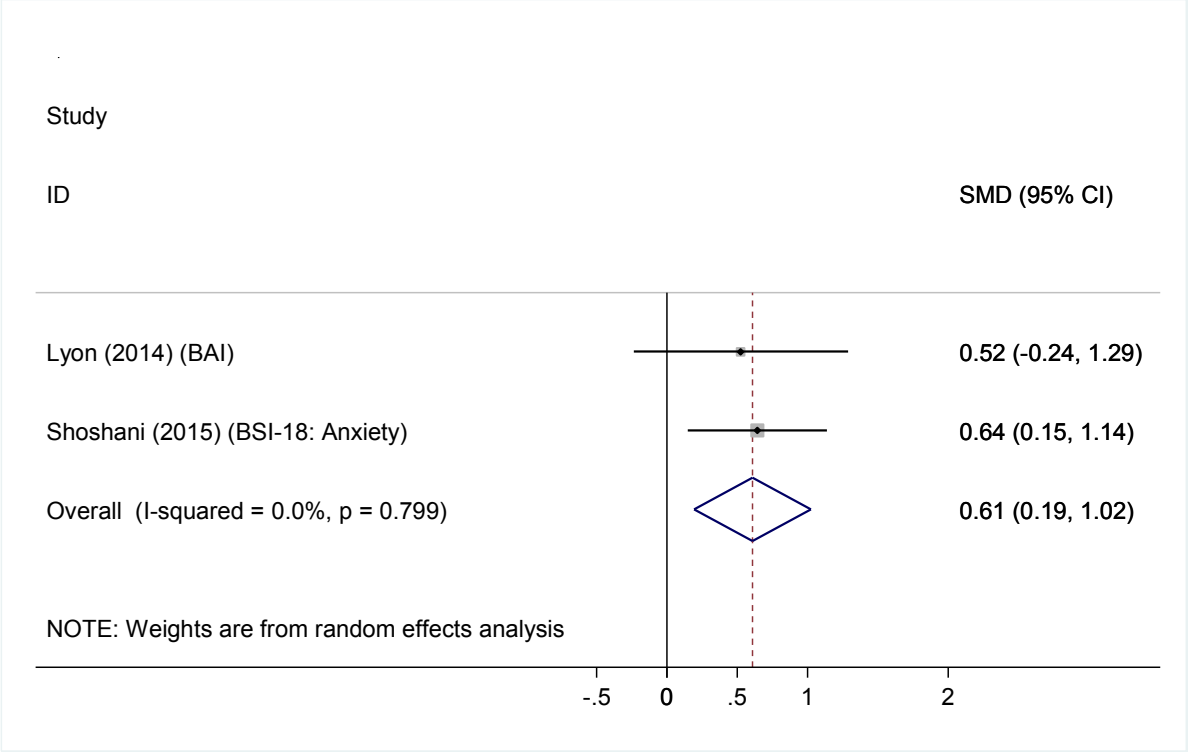

Figure 9: Forest plot showing the results of meta-analysis of the effects of palliative care on child anxiety at post-intervention for CYP with cancer. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; BAI = Beck Anxiety Inventory; Brief Symptom Inventory-18 item version.

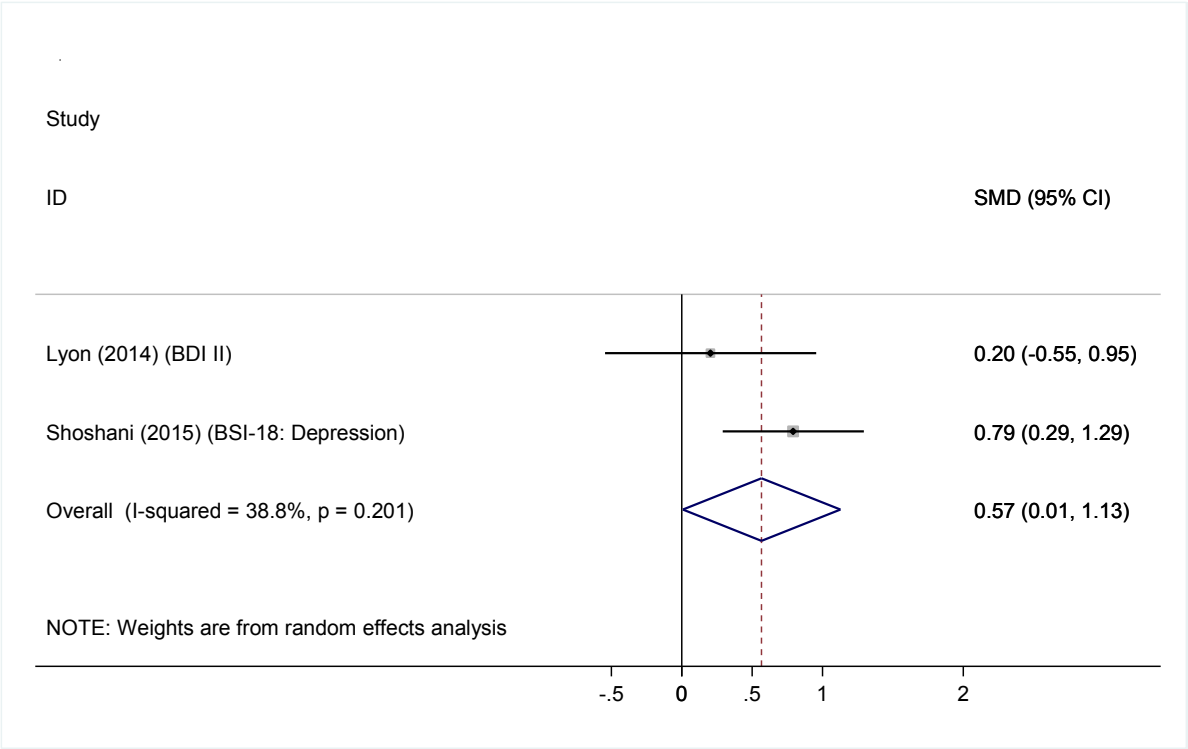

Figure 10: Forest plot showing the results of meta-analysis of the effects of palliative care on child depression at post-intervention for CYP with cancer. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; BDI II = Beck Depression Inventory 2; Brief Symptom Inventory-18 item version.

Other outcomes

*Supplementary Materials 1, Table 14*displays findings for each other outcome assessed at post-intervention. There was little evidence for the beneficial effect of FACE-TC for the majority of outcomes reported. However, Lyon and colleagues did find a large beneficial effect for spirituality (d = 0.99; 95% CI: 0.19 to 1.78, p = 0.02), albeit with imprecision in the estimate of effect with the confidence interval including small effect sizes.[122] Shoshani and colleagues provide evidence of a large beneficial effect for the Make a Wish intervention for physical quality of life (d = 0.87; 95% CI: 0.37 to 1.38, p < 0.001).[136]

Both studies measured intervention effects using the PedsQL 4.0: Physical functioning subscale and therefore physical quality of life specifically could be meta-analysed across the two studies (see *Figure 11*). The result of this meta-analysis provides evidence for a medium sized beneficial effect of these palliative care interventions on physical aspects of quality of life in CYP with cancer (d = 0.73; 95% CI: 0.31 to 1.15, p = 0.001). As both studies used the PedsQL 4.0: Physical functioning subscale, meta-analysis using mean differences can be seen in *Appendix 6*.

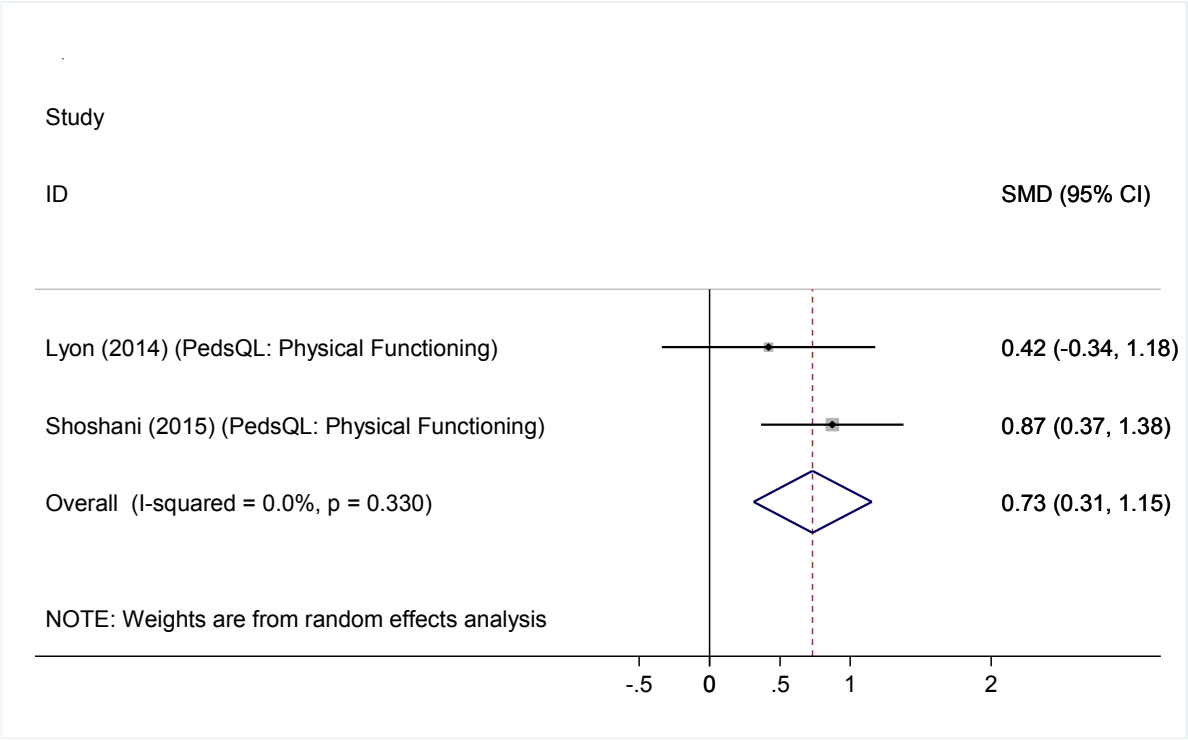

Figure 11: Forest plot showing the results of meta-analysis of the effects of palliative care on child physical quality of life at post-intervention for CYP with cancer. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; PedsQL – Pediatric Quality of Life Inventory.

Overall, analysis of the effectiveness of palliative care interventions for CYP with cancer suggests tentative evidence that there may be some benefits for child depression and anxiety, child physical quality of life and child spirituality. Only two studies investigated the effectiveness of this type of intervention, with differences in findings perhaps reflecting the different intervention content.[122, 136] The relatively low quality of the two articles further limits the confidence of evidence regarding whether palliative care interventions show beneficial effects for children with mental ill health and cancer.

Effectiveness of relaxation interventions

Two studies evaluated the effectiveness of relaxation interventions, both included children with asthma.[118, 138] The sample studied by Bignall and colleagues had elevated symptoms of anxiety, while the CYP in Yang and colleagues' study had elevated depression and anxiety symptoms. Both interventions assessed included PMR.[118, 138] The study by Yang and colleagues was conducted in China, and assessed 30 minutes per night of audio-taped PMR for four weeks versus a TAU control.[138] The study by Bignall and colleagues was based in the USA, and compared a programme of asthma education, breathing exercises, guided imagery and CD-guided PMR to a control group receiving the asthma education alone.[118] A total of 94 participants were included, of which 47 were randomised to the intervention arms. Both studies aimed to reduce anxiety, with the relaxation intervention trialled by Yang and colleagues also targeting depression.[138] It is notable that the two studies reporting relaxation interventions scored poorly on quality appraisal.

Children and young people ***mental health outcomes***

Supplementary Materials 1, Table 15 displays findings for CYP mental health outcomes assessed at post-intervention. Evidence for the positive effect of PMR is provided by the study by Yang and colleagues (2004), where a large, beneficial effect for depression (d = 1.01; 95%CI: 0.48 to 1.43, $p < 0.001$) was observed.[138] Yang and colleagues also reported a medium, beneficial effect for anxiety (d = 0.70; 95% CI: 0.19 to 1.20, p = 0.007), although the confidence interval for this effect includes some small beneficial effect sizes. In contrast there was little evidence reported by Bignall and colleagues for the effect of their relaxation intervention on anxiety, with small harmful effect sizes reported, although confidence intervals were wide and included positive effects (state anxiety: d = -0.39; 95% CI: -1.11 to 0.34, p = 0.30; trait anxiety: d = -0.21; 95%CI: -0.93 to 0.51, p = 0.58).[118]

Given that the same asthma education appeared in both Bignall and colleagues' intervention and control groups,[118] we meta-analysed the anxiety outcomes alongside the data from Yang and colleagues' study,[138] whose control group was TAU (see *Figure 12*). The Trait anxiety subscale of the STAI was more aligned with the SCARED assessment tool, therefore these outcomes were meta-analysed, rather than including the state subscale data from Bignall and colleagues.[118] Meta-analysis of these outcomes provides a lack of evidence for the effect of relaxation interventions on anxiety (d = 0.28; 95% CI: -0.60 to 1.16, p = 0.53).

Heterogeneity across the two papers was wide (I^2 = 75.3%), further impacting the confidence in the pooled effect.

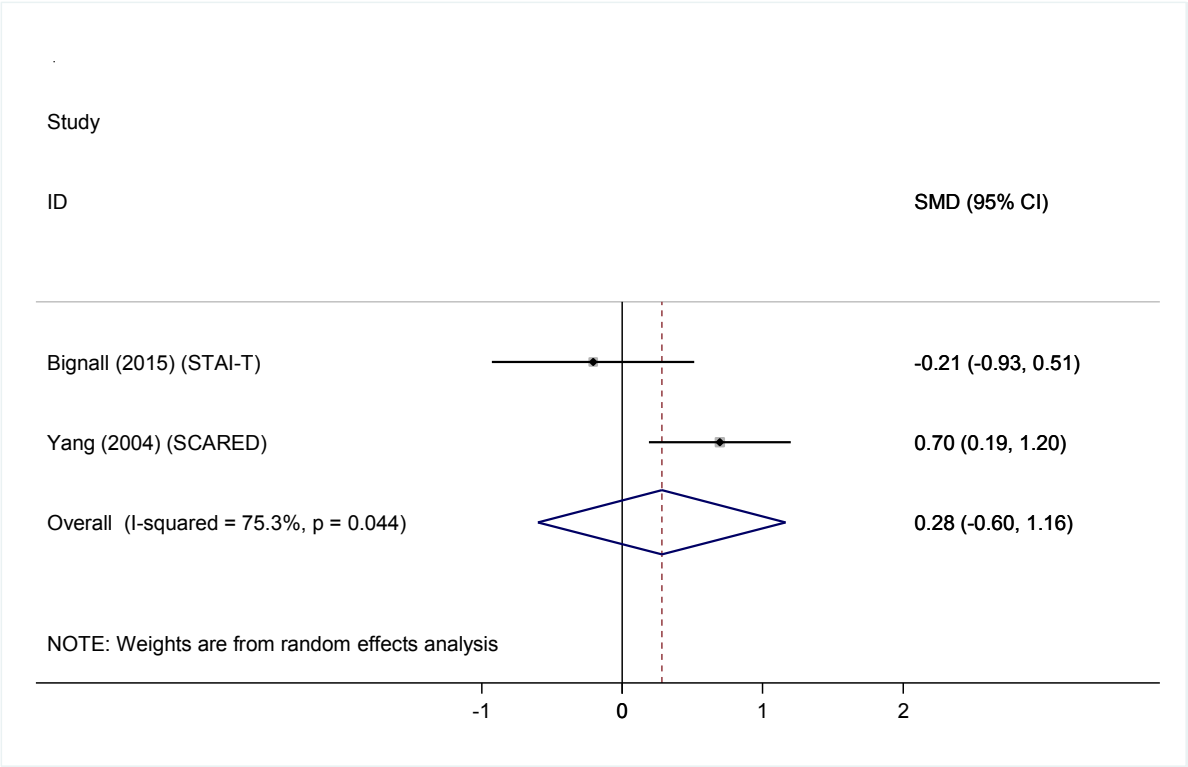

Figure 12: Forest plot showing the results of meta-analysis of the effects of relaxation on child anxiety at post-intervention for CYP with asthma. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; STAI-T = Spielberger State-Trait Anxiety Index – Trait subscale; SCARED = Screen for Child Anxiety Related Emotion Disorder.

Other outcomes

Both studies included assessments of asthma symptoms. Yang and colleagues recorded the incidence of daytime and night time symptoms, as well as the percentages of days without asthma symptoms and of participants experiencing one or more asthma attacks.[138] However, these outcomes were not reported in terms of means and SDs, therefore were not included in data synthesis. Additionally, Bignall and colleagues included a measure of LTC-specific quality of life, the PedsQL: Asthma module.[118]

Supplementary Materials 1, Table 16 displays findings for other outcomes assessed at post-intervention. Evidence for the effect of PMR on asthma symptoms is provided by Yang and colleagues, with large beneficial effects on the number of daytime (d = 2.32; 95% CI: 1.68 to 2.96, $p < 0.001$) and night time (d = 1.91; 95% CI: 1.31 to 2.50, $p < 0.001$) symptoms reported.[138] However, this was only corroborated by one measure of asthma symptoms in Bignall and colleagues, where a large effect size was found for asthma control, although the wide confidence intervals included some negligible effect sizes (d = 0.80; 95% CI: 0.05 to

1.54, $p = 0.04$).[118] There was a lack of evidence from the study by Bignall and colleagues for other measures of asthma symptoms. For asthma-specific quality of life there was a medium effect size, although this estimate of effect is uncertain with a wide confidence interval including negligible effect sizes.[118]

Combined LTC symptom outcomes were produced for each study and entered into a meta-analysis (see *Figure 13*), which showed a lack of evidence for the effect of relaxation interventions on LTC symptoms. Despite a large pooled effect, it was characterised by very wide confidence intervals including harmful effects as well as very large heterogeneity (I^2 = 97.4%) on account of the differences between each study's findings (d = 1.10; 95% CI: -0.88 to 3.08, p = 0.28).

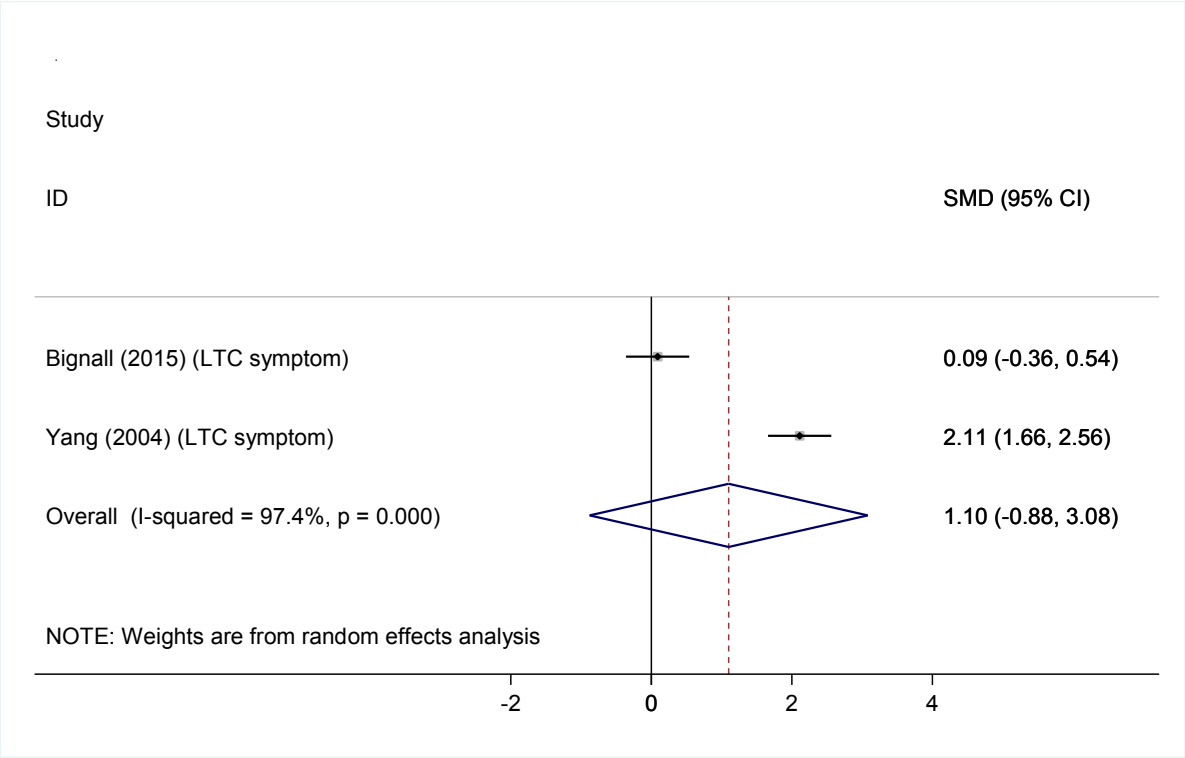

Figure 13: Forest plot showing the results of meta-analysis of the effects of relaxation on asthma symptoms at post-intervention for included studies. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; LTC = Long term condition.

Overall, the included studies provide mixed evidence regarding the benefits of relaxation interventions for depression, anxiety and LTC symptoms. Yang and colleagues reported medium to large beneficial effects across these domains.[138] Bignall and colleagues reported little evidence for the beneficial effects of a combined relaxation intervention.[118] A reason for this may be the asthma education active comparator group in the trial by Bignall and colleagues, which did improve asthma symptoms compared to baseline.[118] Both studies were of low quality/high risk of bias. We recommend the positive effects from Yang and colleagues be treated with caution.

Effectiveness of stress management training interventions

Two studies conducted in the USA evaluated the effectiveness of stress management training (SMT) interventions in children with T1DM and elevated stress[119] and anxiety.[121] Hains and colleagues structured a SMT programme over six weekly one hour sessions,[121] while Boardway and colleagues began with ten sessions of SMT delivered over 12 weeks, followed by three monthly sessions focused on dietary compliance and insulin administration.[119] The

SMT trialled by Hains and colleagues was described as a cognitive-behaviour stress inoculation training programme, in that there was an emphasis on identifying potential illness-related stressors and thinking about them in a more positive way.[121] Boardway and colleagues used a somewhat similar approach, but developed problem solving or coping activities in response to identified stressors.[119] Fifteen participants received SMT, 16 control participants received TAU, with Hains and colleagues employing a waitlist design.[121] Both studies targeted diabetes-specific stress as their primary outcome. The included studies are the oldest in this review, and both scored relatively poorly on quality appraisal.

Children and young people ***mental health outcomes***

Supplementary Materials 1, Table 17 displays findings for each CYP mental health outcome assessed at post-intervention. There was little evidence across both studies regarding the effectiveness of stress management programs on CYP mental health outcomes. On the occasions where medium sized effects were recorded, these were characterised by wide 95% confidence intervals that include harmful effect sizes. This was the case for state anxiety (d = 0.69; 95% CI: -0.41 to 1.78, p = 0.23) and behavioural coping in the study by Hains and colleagues (d = 0.65; 95% CI: -0.44 to 1.74, p = 0.25).[121]

Meta-analysis was possible for their LTC-specific stress and coping outcomes. *Figure 14* shows a forest plot for the effects of SMT on diabetes-specific stress at post-intervention. Meta-analysis revealed little evidence of a beneficial effect for LTC-specific stress (d = 0.10; 95% CI: -0.62 to 0.82, p = 0.80). As both studies used the DSQ, meta-analysis using mean differences can be seen in *Appendix 6*. *Figure 15* shows a forest plot for the effects of SMT on coping at post-intervention reported by Boardway and colleagues and Hains and colleagues.[119, 121] Negative coping from the Kidcope measure was selected from Hains and colleagues as this subscale has most similarities with Boardway and colleagues' Ways of Coping Checklist. Meta-analysis again revealed little evidence for an effect of the intervention on coping (d = -0.09; 95% CI: -0.83 to 0.64, p = 0.81).

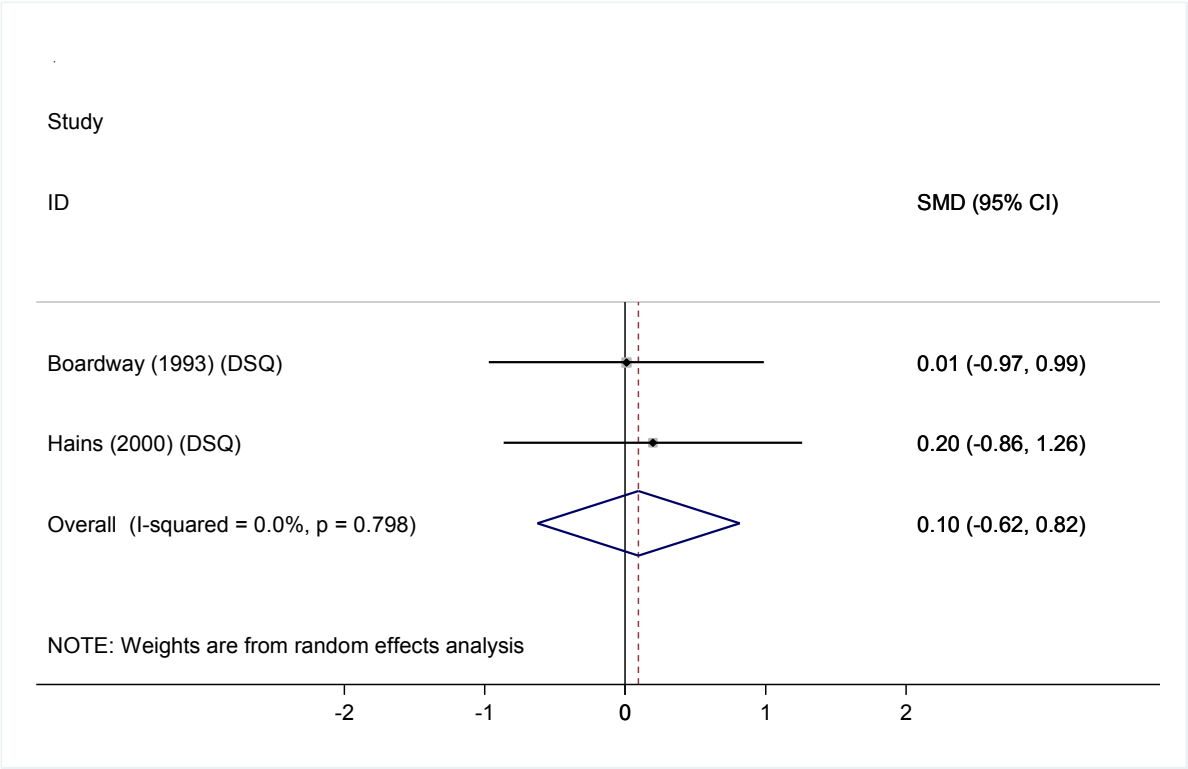

Figure 14: Forest plot showing the results of meta-analysis of the effects of stress management training on T1DM-specific stress at post-intervention for CYP with diabetes. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval; DSQ = Diabetes Stress Questionnaire.

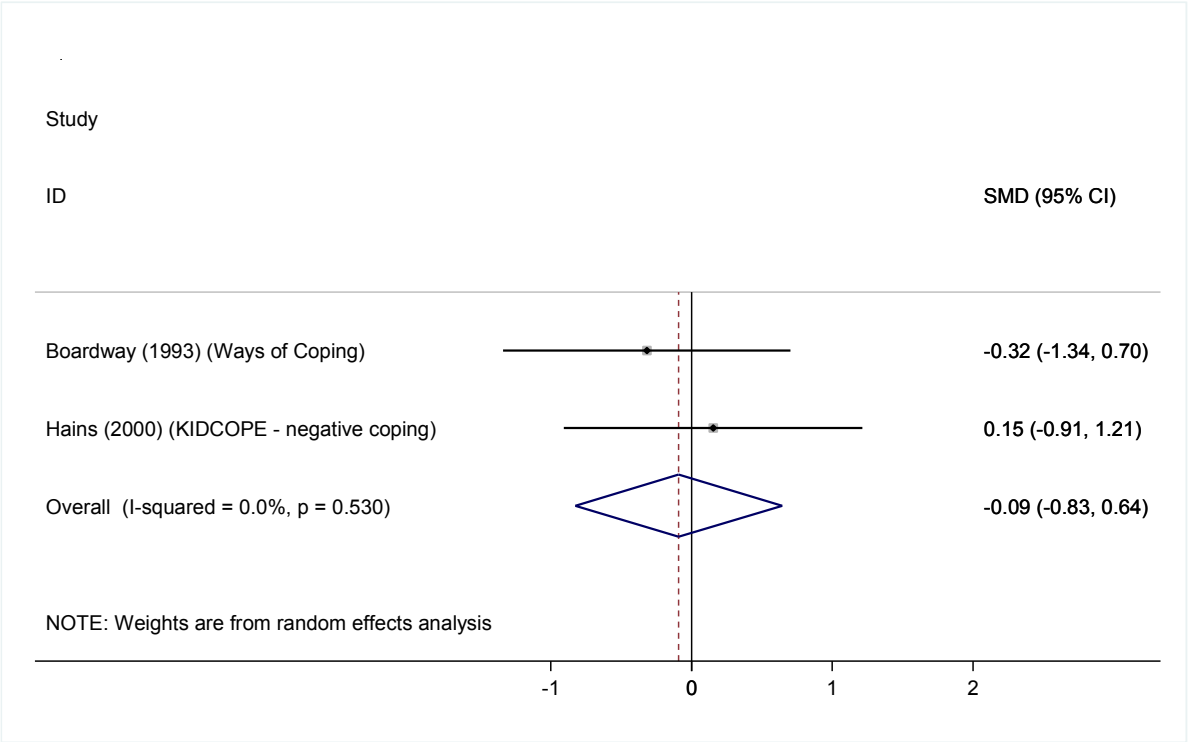

Figure 15: Forest plot showing the results of meta-analysis of the effects of stress management training on coping at post-intervention for CYP with diabetes. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval.

Other outcomes

Supplementary Materials 1, Table 18 displays findings for each other outcome assessed at post-intervention. There was again a lack of evidence for the effect of stress management interventions on any outcome. LTC symptoms were meta-analysed for the two studies using the most similar glycaemic control outcomes (GHb in Boardway et al.,[119] HbA1C in Hains et al.[121]). This meta-analysis (see *Figure 16*) provides little evidence for the effect of stress management training on LTC symptoms (d = -0.10; 95% CI: -1.77 to 1.57, p = 0.91), showing imprecise effects owing to a wide confidence interval and large heterogeneity across the two studies (I^2 = 79.9%).

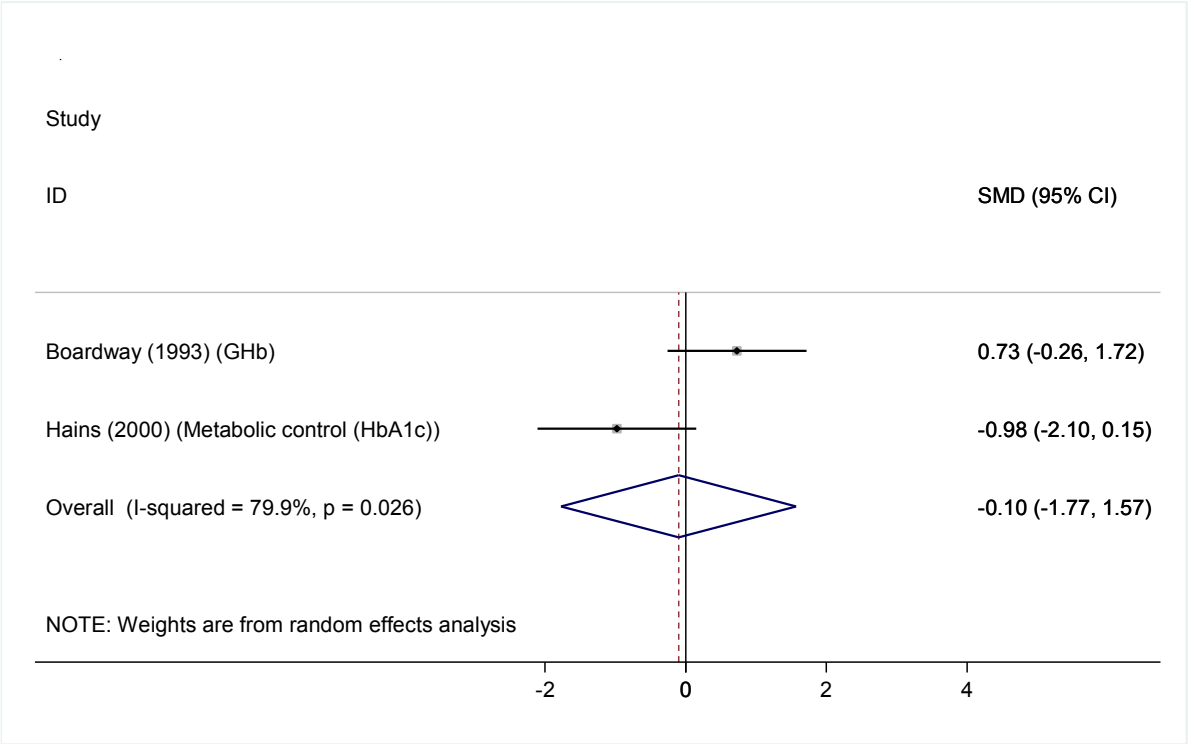

Figure 16: Forest plot showing the results of meta-analysis of the effects of stress management training on CYP diabetes symptoms at post-intervention. SMD = Standardised Mean Difference (Cohen's *d*); CI = Confidence Interval.

Overall, the two included studies provide very little evidence regarding the effect of stress management training on a wide range of outcomes. Effect sizes ranged from medium sized and beneficial to large harmful effects, but all were characterised by imprecision in the estimate with large effect sizes, at least in part due to the small sample sizes in these studies.[119, 121]

Effectiveness of emotional intelligence training interventions

Two studies based in Iran evaluated the effectiveness of emotional intelligence training (EIT) in children with hearing loss and elevated distress and general mental health symptoms.[131, 133] The article by Ashori and colleagues was translated from Persian; it tested a programme based on nine modules related to understanding and controlling emotions,[131] while the study by Pourmohamadreza-Tajrishi and colleagues featured 11 similar modules.[133] Each study contained 40 participants recruited from specialist schools for the deaf, equally split between EIT and TAU. The two included studies were of very poor quality.

Children and young people ***mental health outcomes***

Both EIT studies used the 28-item version of the General Health Questionnaire (GHQ-28) and reported subscales for anxiety, depression, social dysfunction, somatic symptoms and overall mental health. There were no other outcomes reported.

Supplementary Materials 1, Table 19 displays findings for these study outcomes. Both studies reported very large, positive beneficial effects for each outcome domain. Effect sizes ranged from 2.41 (95% CI: 1.58 to 3.23, $p < 0.001$) for depression to 6.79 (95% CI: 5.14 to 8.44, $p < 0.001$) for overall mental health in the study by Pourmohamadreza-Tajrishi and colleagues,[133] while Ashori and colleagues reported effect sizes in the range 11.0 to 17.4 for the same outcomes.[131]

Initial assessment of the two papers suggested that they were reporting on different studies, due to differences in the EIT programme outline and age of the sample. Upon inspection of the raw data presented for each study, the review team decided that the mean scores were too similar to be confident that these were in fact different samples, and therefore meta-analysis was not performed. It is also noted that extremely large effect sizes are evident due in part to the very small standard deviations presented. We attempted to contact the authors for clarification but received no reply.

Despite the reports of very large beneficial effects in the individual studies of Ashori and colleagues and Pourmohamadreza-Tajrishi and colleagues, both studies scored poorly on quality appraisal, notwithstanding the conspicuous similarities between the studies.[131, 133] Overall, there is a lack of evidence to confidently determine the effect of EIT on the mental health of CYP with hearing loss.

Effectiveness of massage therapy interventions

The study by Diego and colleagues was the only one to assess a massage therapy intervention, studying its effects in adolescents with Human Immunodeficiency Virus (HIV) and symptoms of depression and anxiety. The study hypothesised that HIV symptoms, anxiety and depression would be improved in the massage therapy group compared to the PMR control group after 12 weeks of the intervention. Twenty four adolescents were randomised equally to massage therapy or relaxation involving PMR.[120]

Children and young people ***mental health outcomes***

Diego and colleagues assessed anxiety using the STAI-S and depression using the Centre for Epidemiological Studies-Depression scale.[120] *Supplementary Materials 1, Table 20* displays these findings. There is evidence that massage therapy had a beneficial effect on child anxiety, with a large, positive effect size calculated (d = 1.23; 95% CI: 0.35 to 2.10, p = 0.007). Despite a medium positive effect size for depression, there is a lack of evidence for this beneficial effect, given the wide confidence interval that includes negligible effect sizes (d = 0.75; 95% CI: -0.08 to 1.58, p = 0.08).

Other outcomes

Diego and colleagues reported the effects of Massage Therapy on markers of immune function.[120] Medical charts were reviewed to assess HIV progression (CD4 cells per mm^3, CD8 cells per mm^3, CD4:CD8 ratio), and protection against viruses associated with HIV (CD56 cells per mm^3, CD56:CD3 ratio). Overall there is a lack of evidence for the effects of massage therapy on HIV symptoms.

The evidence provided in the study by Diego and colleagues suggests that massage therapy may reduce anxiety and to a lesser extent depression in adolescents with HIV.[120] However, there was a lack of evidence for effects on markers of immune function. This was the only study reporting massage therapy, and it was of low quality, therefore there is a lack of unequivocal evidence regarding the effects of this intervention on the mental health of CYP with LTCs. It is notable that the comparator group in this study was similar to components of interventions tested in the studies by Bignall and colleagues[118] and Yang and colleagues[138] which were earlier categorised as relaxation interventions.

Effectiveness of heart-rate-variability biofeedback interventions

Yetwin was the only included study to assess a heart-rate-variability biofeedback (HRVB) intervention, studying its effects in a sample of 19 adolescents with chronic pain and elevated anxiety symptoms.[126] The nine participants in the intervention group practised the use of breathing and relaxation to control their heart rate, while linked to a heart rate monitor. The control group was enrolled in a waitlist design.

Children and young people mental health outcomes

Child anxiety and depression were self-reported using the Children's Anxiety Sensitivity Index and Child Depression Inventory-Short form respectively. *Supplementary Materials 1, Table 21*displays findings for these outcomes as measured at post-intervention. There was little evidence for the effect of HRVB on either anxiety or depression.

Other Outcomes

Yetwin assessed quality of life, quality of sleep and LTC symptoms (pain).[126] *Supplementary Materials 1, Table 22* displays findings for these other outcomes measured at post-intervention. There were a couple of medium to large effect sizes reported for both LTC symptom and sleep outcomes; however, these were characterised by imprecision in the estimates, with confidence intervals including negligible effect sizes (e.g. Faces Pain Scale: d = 0.86; 95% CI: -0.09 to 1.81, p = 0.08 and Child/Adolescent Sleep-Wake Scale -Falling asleep: d = 0.78; 95% CI: -0.16 to 1.72, p = 0.11). This tentative evidence of beneficial effect also conflicted with other measures of these outcomes that showed a lack of evidence of beneficial effects (e.g. Varni-Thompson Pediatric Pain Questionnaire Worst pain: d = -0.05; 95% CI: -0.95 to 0.85, p = 0.92 and C/ASWS Sleep Total Score: d = 0.27; 95%CI: -0.63 to 1.18, p = 0.56).

Overall, the one paper assessing HRVB provides a lack of evidence for its effect on child anxiety, depression, sleep, quality of life or pain in adolescents with chronic pain. In addition to being the sole study to use this type of intervention, Yetwin scored very poorly on quality appraisal; therefore there is insufficient evidence to assess the effectiveness of HRVB for improving either mental health or other outcomes in CYP with LTCs.[126]

Effectiveness of resistance training interventions

One study investigated the influence of a resistance training programme on depression and physical symptoms in a sample of 22 adolescents with chronic fatigue syndrome.[128] The programme was progressive (gradually increasing in difficulty), utilising exercises such as weight lifting and sit-ups, and was compared to a programme consisting of aerobic exercises alone.

Children and young people mental health outcomes

Depression was self-reported using the Beck Depression Inventory. *Supplementary Materials 1, Table 23* displays the findings for this measure and other measures described below at post-intervention. There was a lack of evidence for the effect of resistance training on depression (d = -0.23; 95% CI: -1.07 to 0.60, p = 0.59) compared with an aerobic exercise programme.

Other outcomes

Gordon and colleagues assessed quality of life using the Physical subscale of the Short Form 36 Health Survey (SF-36).[128] LTC symptoms were measured using the Fatigue Severity Scale, and several exercise tests chosen to assess exercise tolerance and muscular endurance including: the number of push ups completed until fatigue; the sit-to-stand test; time to fatigue on a submaximal walking test and metabolic equivalent during the walking test (ratio of energy expenditure during the test to energy expenditure at rest). There was a lack of evidence for the effects of resistance training on these outcomes.

Overall, the one paper assessing the effects of a resistance training programme on depression, physical indices and quality of life, provides little evidence of a benefit compared to aerobic training. It should be noted that Gordon and colleagues scored moderately well on quality appraisal, describing randomisation, allocation concealment, blinding of outcome assessors and using an intention to treat analysis. However, groups were imbalanced at baseline, where the resistance training group had performed statistically significantly better on the sit-to-stand test, and this was not adjusted for; the intervention was not well described and adherence, compliance and fidelity were not assessed. As such, there is insufficient evidence to state with confidence whether resistance training has any benefit for the mental health of CYP with LTCs.

Effectiveness of music therapy interventions

Bufalini was the only included study to assess a music therapy intervention.[135] It also differed from other included studies as it was written in Italian and aimed to alleviate procedural anxiety, rather than general anxiety. It included 39 adolescents with cancer undergoing painful procedures in Italy. The intervention consisted of actively listening to music, playing instruments, and then having background music playing prior to sedation. The 19 participants randomised to the control group received usual sedation.

Children and young people mental health outcomes

Bufalini assessed anxiety at four time points using the modified Yale Preoperative Anxiety Scale (mYPAS).[135] *Supplementary Materials 1, Table 24* displays findings at post-intervention, just prior to sedation. There is evidence that music therapy had a large, beneficial effect on child anxiety (d = 1.47; 95% CI: 0.76 to 2.18, $p < 0.001$). No other outcomes were measured in this study.

The evidence provided by Bufalini suggests there may be a benefit of music therapy in reducing procedural anxiety.[135] However, in addition to being the sole paper reporting on this intervention, it scored the lowest of all included studies on quality appraisal. Therefore, more evidence is required to increase confidence in the effectiveness of music therapy for improving the mental health of CYP with LTCs.

Discussion

Summary of findings

This review synthesised RCTs studying the effectiveness of interventions which aim to reduce the mental ill health of CYP with LTCs. The primary focus of the review is the mental health of CYP; yet all secondary outcomes were synthesised in order to evaluate any wider benefits of interventions, including key aspects of individual and family functioning.

Of the 11 reported intervention types, the greatest volume of research focussed on the effectiveness of CBT, with seven studies evaluating CBT-based interventions. These studies provide tentative evidence that CBT-based interventions could be beneficial for the mental health of CYP with LTCs, with six of seven studies of CBT interventions reporting improvements in CYP mental health outcomes. However, the heterogeneity of the included study characteristics (comparators, LTC and outcomes) meant that no meta-analyses could be conducted for CYP mental health outcomes in our main analysis, leading to a reliance on individual study outcomes.

Parenting programmes featured in four interventions, tested in three studies.[117, 127, 130] Two interventions included ACT with the SSTP parenting programme and there appeared to be evidence of beneficial effects of this combined programme for child behaviour, emotional problems, parenting and parent mental health compared to SSTP alone. Group Play Therapy interventions were assessed in three studies[132, 134, 137]. Quality appraisal indicated a high risk of bias in each study and where large positive effect sizes were seen, these were all characterised by uncertainty about the true effect of these interventions. It should be noted that reliance on parent-reported outcomes introduced further potential for bias when assessing parenting programmes.

The effects of stress management training,[119, 121] emotional intelligence training,[131, 133] palliative care[122, 140,136] and relaxation interventions[118, 138] were each reported in two studies with four further interventions reported in single studies (biofeedback,[126] music therapy,[135] resistance training[128] and massage therapy[120]). While there were some beneficial effects reported for emotional intelligence training, palliative care and massage therapy, risk of bias in these studies limits the conclusions that can be drawn regarding these interventions.

The 25 included studies reported on 269 individual outcomes; however, there were relatively few opportunities to meta-analyse studies with similar intervention, comparator, LTC and outcomes. No more than two studies shared particular combinations of these study characteristics and could be meta-analysed in the main analysis. Even when we considered pooling studies across LTCs for

supplementary analyses, this only allowed for further pooling of CBT interventions (see *Appendix 7*). Trials were typically small, with only two including more than 100 CYP participants.[125, 129] Effect sizes across the board were therefore characterised by wide confidence intervals, meaning that on a range of occasions where calculation of standardised mean differences indicated medium or large effects, these estimates were consequently imprecise. The evidence for particular interventions used with similar samples of CYP is very limited.

Results in context

Despite the high prevalence of CYP with LTCs and comorbid mental illness, this review highlights the lack of randomised controlled trials aiming to address the problem. It is notable that 21 of the 31 included papers were published since 2010, 13 since 2014, perhaps suggesting that this lack of research is beginning to be addressed. However, study quality of the relatively small amount of research to date is poor, with 19 of the 25 studies having at least four separate indicators of risk of bias.

The suggestion that there may be promise for CBT in improving the mental health of CYP with LTCs supports existing evidence. The most relevant previous review, by Bennett and colleagues[81] published a systematic review of psychological interventions for symptoms of mental health disorders in CYP with LTCs, finding preliminary evidence that CBT interventions may be beneficial. These authors located only two RCTs,[82, 83] both of which appear in the current review, and both of which reported beneficial effects for depression outcomes. Bennett and colleagues required the presence of symptoms of a DSM-IV or DSM-5 mental health disorder for inclusion in their review,[81] and had narrower inclusion criteria regarding LTCs and mental health outcome measures than the present review. In addition to those studies identified by Bennett and colleagues,[81] we found two RCTs published since that review which included participants with a diagnosed mental health disorder: Reigada and colleagues[124], principal anxiety disorder) and Szigethy and colleagues,[125] major or minor depression diagnosis). As highlighted previously, ours is the first systematic review to consider all treatments for elevated symptoms of mental ill health in CYP with LTCs. By examining this literature, our findings add to the evidence from the small number of trials examining samples with diagnosed mental health disorders. Extending the criteria to include CYP with clinically significant levels of mental health difficulties, as well as those with a mental health diagnosis, enabled a wider range of studies to be included and acknowledges the significant impact on children experiencing some psychological difficulties associated with their LTC.

Wider evidence of the effectiveness of CBT for improving mental health has informed NICE guidelines recommending CBT for CYP with depression.[150] James and colleagues[69] published a Cochrane systematic review of CBT for childhood anxiety disorders in CYP without comorbid LTCs, reporting that, despite a limited evidence base, there was a suggestion that CBT is a more effective treatment than TAU or active comparators. Given our own findings, there is tentative evidence that this recommendation may hold for CYP with LTCs.

It was observed that five of the six CBT interventions included in this review had been adapted prior to delivery to contain LTC-specific content relevant to the CYP in the study.[83, 123-125, 129, 145] Content was adapted to the extent that the intervention would only be suitable for a sample with that LTC. Of these studies, beneficial effects on CYP mental health outcomes were observed following four of the interventions, with only Best of Coping failing to observe any benefits.[129] Of the other interventions types evaluated in studies included in this review, only two others were adapted to the LTC, with neither showing evidence of benefit CYP mental health outcomes.[118, 119] The mixed evidence for interventions which are adapted to the LTC means it is unclear how this should be done to best effect. There was a lack of evidence located that had assessed the effects of existing non-adapted CBT that has not been modified or adapted to the LTC in populations with an LTC. Although Martinović and colleagues demonstrated some positive effects of CBT for depression in a sample with epilepsy, there were issues with study quality,[82] indicating a gap for RCTs assessing how well CBT interventions that have been shown to be effective in physically well CYP populations work for CYP with LTCs.

The strengths and limitations of the review, and the implications and recommendations for future work are discussed in Chapter 6.

Chapter 3 Review 2: Experience of interventions targeting mental health for children and young people with long term physical conditions

Research questions

This chapter describes the second systematic review and addresses the following research questions:

1) What are the perceived effects of interventions aiming to improve mental health and wellbeing for children and young people (CYP) with long-term physical conditions (LTCs) on mental health and other key aspects of individual and family functioning?

2) What are the factors that may enhance, or hinder, the effectiveness of interventions and / or the successful implementation of interventions intended to improve mental health and wellbeing for CYP with LTCs?

For definitions of the key terms used within this chapter, please see Chapter 1

Methods

The steps used to identify and select evidence were as for Review 1.

Identification of evidence

The inclusion criteria shown in Supplementary Materials 2, Table 1 were agreed amongst the wider project team, and used to determine eligibility of articles. The inclusion criteria specified primary qualitative studies that explore attitudes and experiences regarding interventions aiming to improve mental health and wellbeing for CYP with LTCs from the perspectives of CYP with LTCs, their families and/or practitioners. Articles did not have to report elevated symptoms of mental ill-health for participants at baseline to be eligible for inclusion, as was the case for Review 1. Due to the nature of qualitative research, it was anticipated that it would have been unlikely that mental health symptoms would have been measured. This would have severely limited the number of studies eligible for inclusion in our review, as highlighted within the '*Quality Appraisal*' section below.

Search strategy

A search strategy using both controlled (e.g. MeSH) and free-text searching was developed and tested in the databases to be searched. Terms were grouped according to four concepts:

- Children and young people
- Mental health terms
- Long term physical conditions terms
- Study design terms (qualitative research)

CYP and LTC terms were the same as those used for Review 1. Mental health terms included those used for Review 1 as well as additional terms capturing intervention targets informed by discussion with experts amongst the wider project team (See *Appendix 8* for list of project team and advisory group members). Additional mental health terms such as "wellbeing" and "coping" were added to the search strategy to reflect the terminology it was anticipated would be used by authors and participants of studies to be included in the review. These search terms were consistent with the broad definition of mental health used within this linked evidence synthesis (See Chapter 1)

MEDLINE including MEDLINE in-process (via OvidSp), PsycINFO (via OvidSp), and CINAHL (via EBSCOhost) were searched. These databases were selected because of their known coverage of qualitative literature.[157, 158] The other databases from Review 1 were not searched because

of their focus on quantitative literature, but any qualitative studies identified during study selection in Review 1 were marked for possible inclusion in this review. No date restrictions were applied to the search. Searches were conducted on 19th April 2016. An example search strategy used for the MEDLINE database is shown in *Appendix 9*. All references identified by the searches were exported into EndNote X7 prior to screening and eliminating duplication.

Supplementary searches were also conducted. Backward citation chasing (searching the references of included articles) searches were conducted by four reviewers (DM, MN, LS, ZK) to locate further primary articles of potential relevance. Included studies from relevant reviews located during title and abstract screening for both Review 1 and 2 were also checked. Forward citation chasing (searching articles citing included articles using Web of Knowledge) searches were conducted by an information specialist (JTB) using Web of Science and Google Scholar. In addition, DM, LS and MN searched websites that had been identified by the project team and advisory group for relevant research (see *Appendix 3* for a list of websites searched). The databases CINAHL, HMIC and Conference Proceedings Citation Index were searched, all of which index grey literature. We also searched the website OpenGrey via http://www.opengrey.eu/.

Relevant studies were identified in two stages based on the inclusion and exclusion criteria given above. First, two reviewers conducted title/abstract screening independently for each record (eight researchers shared this screening: LS, DM, MN, IR, JTB, JTC, MR, VB). Endnote X7 software was used to perform screening. Disagreements were resolved through discussion between two reviewers, with referral to a third reviewer as necessary (DM, MN, LS). Full texts of records that appeared to meet the inclusion criteria on the basis of titles and abstracts were then obtained wherever possible. Full texts were screened independently by two reviewers (nine researchers shared this screening: DM, LS, MN, JTB, JTC, MR, LL, VB, IR) for inclusion and exclusion. Disagreements were again resolved through discussion between two reviewers, with referral to a third reviewer as necessary (DM, MN, LS).

Methods of analysis/synthesis

Data extraction

A data extraction form was developed and piloted. LS, MN and DM each extracted one article and checked one colleague's extraction, before discussing and amending the form. Data on article details and aims, participants, methods, intervention, summary of findings and study quality were extracted into Microsoft Office Excel 2010 (Microsoft, Redmond, WA, USA) by LS, MN, IR and

DM and checked by either DM or LS. Full articles were uploaded to NVivo (v. 11, QSR International, Warrington, UK) software, which was used to aid coding of the findings and interpretation.

Quality appraisal

Quality appraisal was conducted simultaneously with data extraction using criteria from the Wallace checklist.[159] The checklist includes 13 questions covering research questions, underpinning theory, study design, context, the sample, data collection, analysis, relationships between data and findings, limitations, generalisability and ethics. We used questions regardless of whether they were considered essential or desirable in the checklist and separated questions falling under the same category. The question, 'are the interventions of interest clearly described?' was added in line with a previous review of intervention experience.[160] Questions were assigned a response of 'yes', 'no', 'cannot tell' or 'not applicable' for each article. Quality appraisal decisions were made by two reviewers (LS, MN, IR and DM) and disagreements resolved through discussion. The appraisals were used to evaluate study quality and were not used to exclude articles, our inclusion criteria requiring recognised methods of qualitative data collection and analysis provided a basic quality threshold.

Data analysis and synthesis

Data analysis and synthesis followed the principles of meta-ethnography. This approach was developed by Noblit and Hare[161] and has proved to be a useful method to interpret findings across multiple qualitative studies in health research (e.g.[162-164]). Meta-ethnography aims to create new interpretations through a process of reciprocal translation and refutation across studies. Translation involves establishing a common language of interpretation between or amongst the included studies and refutation involves the explanation of contradictory findings. If applicable a 'line-of-argument' synthesis is created that produces a model that synthesises various reciprocal and refutational relationships into a coherent whole.

A challenge faced was the large quantity of articles and breadth/diversity of interventions, LTCs and participant groups. Noblit and Hare[161] speak of the danger that the inclusion of too many studies may lead to "trite conclusions" (p. 28). This was dealt with by grouping articles on the basis of similarities in participant perspective and level of interpretation in article findings, to create a staged process of synthesis. Articles were grouped according to the quantity of relevant findings available and the level of interpretation used by individual study authors when generating themes from their primary patient data, as judged and agreed by reviewers (LS and DM). Synthesis began using articles that had a greater degree of interpretive findings. Supplementary Materials 2, Table 2

provides examples of interpretative and descriptive themes from primary studies included in this review.

This meant that a smaller number of articles were initially synthesised separately before being combined into an overall synthesis, assuming that there were similarities that allowed some translation of ideas between articles. Different participant perspectives were initially analysed separately as follows: CYP, parents, practitioners, mixed perspectives. Interventions targeting procedural anxiety were initially synthesised separately. This allowed for any potential differences in the experiences of procedural interventions aiming to alleviate anxiety in the short-term and interventions which aim to address CYPs mental health and/or wellbeing over a longer period of time to be identified.

We then used purposive sampling to select further included articles with LTCs, interventions and participant perspectives that had not been represented in the synthesis of articles thus far. Purposive sampling has been used in previous qualitative evidence syntheses to manage the analytical process when many studies are included.[165] We used a form of maximum variation sampling to decide whether studies that included interventions and LTCs not seen in the synthesis thus far ought to be synthesised next[166] and to ensure that LTCs and interventions included in Review 1 were prioritised for inclusion in the current synthesis. Studies were ranked according to the degree to which they met these three criteria. Studies that met these criteria (n = 30) were synthesised, which left 12 articles with descriptive findings related to interventions and LTCs previously included in the synthesis whose findings were compared to the existing synthesis. A summary of the stages of the synthesis and articles included at each stage is provided in Table 2 below.

Table 2: Stage of inclusion for articles included within the synthesis of Review 2

Stage of synthesis	Articles included	Rationale for stage of inclusion in synthesis
1) Synthesis of articles with large amount of interpretative findings	167-177 178-182 183-186	Articles with over two pages of interpretative data relating to experience of or attitude towards an intervention. Provision of a rich source of data to begin process of reciprocal translation.
2) Purposive sampling	118, 187-191	Interventions or LTC seen in Review 1.
	192,193-196,197-203	Types of intervention or LTC not yet seen within the Review 2 synthesis.
	196, 204-213	Interventions focused on procedural distress.

3) Read through of descriptive articles	214-221,222-225	These articles did not meet previous criteria. Checked for data to support or refute existing analysis.

The process used for the meta-ethnography as a whole included:

1. Reading and re-reading the included studies

This initial stage involved the careful reading of all of the included articles in order for reviewers to familiarise themselves with the studies and their findings. This process began in the screening phase and continued through data extraction and analysis.

2. Determining how the studies are related

Following data extraction, structured summaries were created in order to juxtapose the same type of information across articles for consideration of similarities and differences.[164] Information tabulated included study aims, samples, methods, interventions and themes. These summaries included information on participant perspective and whether the intervention targeted procedural anxiety to allow categorising of articles for synthesis. Themes from all included studies presented in their findings sections were also tabulated and colour coded separately to allow for early comparison of themes across different studies.

3. Reciprocal translation of studies

Each article was rated by two reviewers (DM, LS) according to whether the analysis was interpretive or descriptive and if there were more than two pages of findings focused on the experience of the intervention. This facilitated a more in-depth consideration of the quantity and quality of interpretive findings available within each article for analysis. This process identified a smaller number of studies with a large amount of interpretive information on which to focus the initial synthesis.

The reciprocal translation stage involved the extraction of first and second order constructs from the studies identified as having a large amount of interpretive findings available for synthesis. First-order construct data was identified as efforts to make sense of daily experiences, often illustrative participant quotes within studies. Second-order construct data consisted of author analysis and interpretation in included articles. Third-order constructs were our interpretations as reviewers which go further than first and second-order constructs in individual articles.[226] Table 3 displays examples of first, second and third-order constructs contributing towards the analysis. Concept maps showing the relationships between analytical ideas and themes across articles were then developed.

Table 3: Examples of first, second and third-order construct data

First-Order Construct	Second-Order Construct	Third-Order Construct
It was fun like it's never been before, I saw someone who is in control of the situation and isn't afraid of diabetes. If he can do it, I've got to succeed....I'm less afraid because of the project; I met people there with amazing abilities. (CYP quote:[167](p. 474)	The mentees admired their mentors. Numerous mentees reported adopting and emulating parts of their mentors' behaviour regarding diabetes.[167]	Hope and Inspiration: Meeting peers who were managing to live successfully with an LTC provided CYP with a source of hope and inspired CYP that they too could live a fulfilling life.

Initially, articles that focused on young people, parents, or practitioners were synthesised separately. However, as we observed considerable overlap between perspectives, all of the articles with large amounts of interpretive findings were synthesised. We compared the findings and interpretations of each article to the concept map developed to that point, adding new constructs and themes where relevant. NVivo software was used to code findings and interpretations line-by-line in articles against the ideas seen in the concept maps, as well as new ideas. Refutational translation was conducted where any quotes, metaphors, themes and concepts which offered oppositional or counterarguments to current findings were specifically searched for and incorporated into the findings. This process was completed with the articles in the initial synthesis, and repeated with findings and interpretations from studies selected through the purposive sampling process. Where new constructs and themes emerged, studies previously examined were checked to see if they could contribute to these new ideas. Studies evaluating procedural mental health interventions were initially considered separately at this point, but due to the similarity in the concepts across these articles and other articles focusing on broader interventions, they were then included in the reciprocal translation process during this purposive sampling phase. The remaining articles which had neither large amounts of interpretive findings or included in the purposive sampling, were then read to see which constructs and themes were supported by the data and if there was any material which refuted the content of these themes. NVivo was used to code any relevant information from these remaining articles. Feedback and ideas relevant to the synthesis from the CYPAG and parent meetings was considered before the reciprocal translation process began and incorporated throughout the different stages of the synthesis as outlined in ***Error! Reference source not found.***.

4. Synthesising translations/creating a line-of-argument

By considering relationships and overlap amongst the constructs and themes within the synthesis, a line-of-argument was developed. A line-of-argument synthesis attempts to make sense of the synthesis as a whole through the development of a new theory or model of understanding. The line-of-argument consisted of a model that captured how the overarching themes, and the categories that contributed towards them, related to one another and captured the experience of participants with an LTC receiving mental health and wellbeing interventions, their families and the professionals involved in their care.

Results

Study selection

The PRISMA diagram in *Figure 17* summarises study selection. Database searches identified 12,285 records, with a further 1,118 of the total records were identified by other means, including citation chasing, included articles from relevant reviews, websites, searches for sibling articles of those included in Review 1 and author contact. After the removal of duplicates, the titles and abstracts of a total of 10,364 records were screened against inclusion and exclusion criteria. The full texts of 355 records were retrieved for further consideration. Of these, 342 (96 per cent) were successfully retrieved. After full text screening, 295 articles were excluded for reasons provided in *Figure 17*. A list of reasons for the exclusion of each article screened at full text is located in *Supplementary Materials 2, Table 3*.

Three studies were each represented by two articles published about the qualitative findings. [214175183222185, 190] Twenty-two of the 60 included articles were located through additional searching.

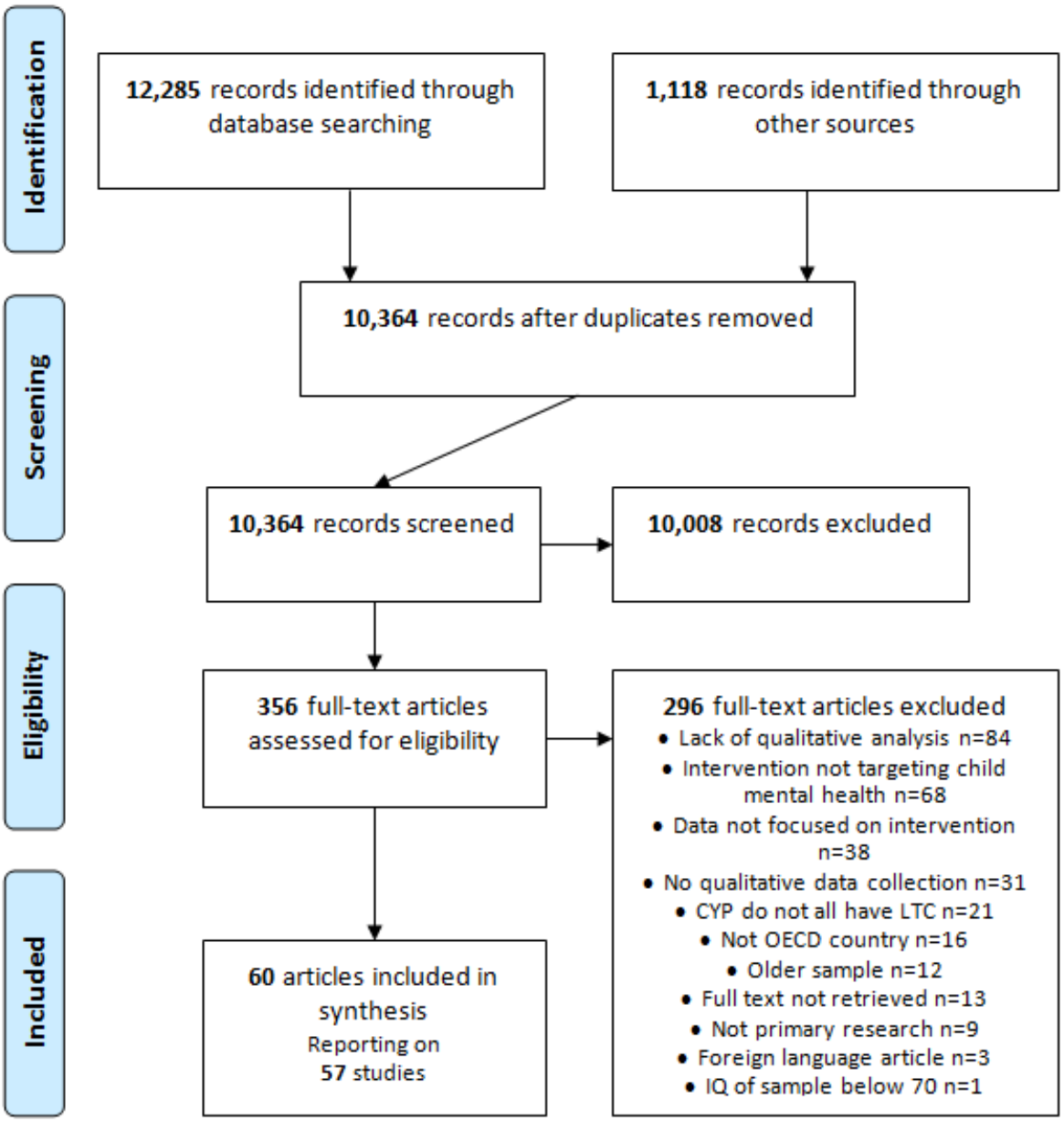

Figure 17: PRISMA flow chart recording Review 2 study selection

Included articles were categorised in terms of the quantity and quality of interpretive findings, participant perspective, intervention and LTC focus. This allowed a stepped approach to synthesis, given the large number of included articles. Twenty articles entered the synthesis at the first stage, which all had a high amount of interpretive findings. Of these, eight reported the views of CYP participants only[167, 168, 170-174, 183], 11 included a mix of participant perspectives[169, 176-184, 186] and two included parent participants only.[170, 185] The different participant perspectives were initially synthesised separately, but considered together on account of the overlap between findings across the articles. In the second stage, 19 articles were selected using a purposive sampling method. Twelve of these articles focused on interventions aiming to alleviate procedural distress.[196, 199, 204-213] These were

initially synthesised separately, but were added to the main synthesis on account of the overlap in ideas which were seen. Finally, the remaining 12 (*See 2*) articles which held descriptive findings were compared to the constructs and themes resulting from the synthesis after the first two stages.

Study characteristics

Supplementary Materials 2, Table 4 provides details about the study location, publication type, focus of the study, method of data collection, method of analysis and intervention type for the 57 included studies. Studies were conducted in seven different countries, with the USA most common (n = 25: [118, 168-173, 177-179, 186, 191, 194-196, 201-203, 206-208, 213, 215, 216, 221]), followed by Canada (n = 13: [174, 181-184, 190, 197, 214, 219, 220, 224, 225 223]), the United Kingdom (n = 9:[176, 187, 188, 192, 193, 200, 204, 217, 218]) and Australia (n = 7:[180, 189, 205, 209-212]). All included articles were published in peer reviewed journals, except for five dissertations, four at PhD level[169, 203, 206, 221] and one at masters level.[190] Three studies were published before 2000, [168, 176, 213] with over three quarters of included studies (76.7 per cent) published since 2010. The majority of studies evaluated an intervention received by CYP with an LTC; ten studies either asked participants about an intervention in development or did this as well as evaluating the intervention once received.[173, 176, 189, 196-198, 202, 217, 224, 225] Two studies relied on participants recollections of interventions previously received,[176, 204] the remainder collected data about current intervention experience.

Twenty-four of the 57 included studies used only qualitative research methods; the remainder also used quantitative methods, although only 20 of these collected quantitative measures of effectiveness alongside qualitative data collection.[118, 169, 175, 180, 186, 191, 194, 195, 198-205, 213, 219, 220] Forty-eight of the included studies involved interviews. The remainder either used focus groups,[173, 189, 202, 211, 217] online posts in discussion forums made as part of the intervention[188, 216, 217, 221] and/or observations.[203] The median sample size was 23 participants (range 3-100); only 18 studies included more than 30 participants.[169, 175, 177, 183, 188, 192, 194, 198, 206, 209, 212, 214, 217, 221-225]

Twenty of the 57 studies included only CYP participants[118, 167, 168, 171-174, 189, 191, 192, 199, 200, 202, 203, 210, 213, 216, 217, 219, 221]. One study collected data from practitioners who delivered the intervention only.[211] Four studies collected data from parents only;[190, 207, 208, 212] meaning the remaining 29 studies included a mix of participant types[169, 170, 176-184, 186-188, 193-198, 204-206, 209, 215, 218, 220, 223, 224]. The most commonly studied intervention types were:

- online support interventions (n = 12: [175, 182, 188, 197, 202, 216, 217, 219, 221, 223-225]),
- camps (n = 9: [168, 170-172, 174, 178, 179, 184, 190]) and
- music therapy (n = 6: [205, 207-211]).

Qualitative data analysis methods typically involved thematic analysis (22 / 57 studies). In 15 studies the authors described the analysis as based on grounded theory or used constant comparative methods.[167, 177-179, 184, 186, 188, 193, 201, 205, 209-211, 213, 223]In nine studies the authors reported that they used some form of qualitative content analysis only.[194, 195, 198, 199, 202, 206, 216, 219, 224]

Few details about populations studied were consistently provided. Over half of the 57 included studies did not report the ethnicity of participants providing qualitative data (n = 31)[167, 168, 170, 171, 176, 178, 182-184, 188, 189, 191, 194, 197-200, 205, 207, 209-211, 214, 217-221, 223-225] and only ten studies provided socioeconomic information about participants or their families.[174, 183, 185, 195, 196, 203), 204, 215, 220, 224] None of the included studies reported that CYP with LTC had received a mental health diagnosis. One study, which was also included in Review 1, indicated that participants had clinically significant levels of anxiety.[118] The study by Curle and colleagues[193] included CYP showing signs of emotional distress related to their LTC.Aside from these two examples, the mental health of CYP participants was not assessed on entry into the study. CYP with an LTC who provided qualitative data were between 8 and 17 years of age in 25 studies.[118, 167, 170-172, 174, 176, 180, 184, 189, 191, 192, 194, 195, 197, 198, 202, 206, 213, 214, 218, 219, 221, 224, 225] Only three studies focused solely on the views of young adults (18 years old or above), two of which were conducted in young adults living with HIV in the USA.[173, 210, 216]

The most frequently seen LTC in CYP in the 57 included studies was cancer (n = 14: [168, 178, 196, 199, 205-213, 217]). HIV positive intervention recipients were the focus of eight studies,[173, 177, 179, 181, 192, 201, 215, 216] while a further ten studies included CYP with a mix of LTCs.[169, 172, 175, 180, 182, 193, 197, 213, 214, 223] Five studies focused on CYP with type 1 diabetes mellitus (T1DM).[167, 189, 195, 202, 219] Included studies commonly explored the perceptions and experiences of interventions aimed at improving coping (n = 26 [168, 171, 179, 182, 189, 191, 192:Curle, 2005 #58, 194:Gaysynsky, 2015 #33, 195-197, 198:O'Callaghan, 2011 #66, 205-208, 213, 214, 217, 219, 220, 222, 223]), self-esteem (n = 13[168, 170, 171, 178, 180, 184, 186, 206, 210, 215-218]) and emotional support (n = 11[176-178, 187, 188, 192, 194, 214-217]) rather than targeting mental health symptoms e.g.

depression (n = 1: [181] or anxiety (n = 8:[118, 170, 181, 182, 201, 203, 204, 212]). Some interventions also aimed to improve symptoms related to the LTC (n = 35[118, 167, 171-173, 176, 177, 179, 181, 182, 186-189, 191, 192, 195, 198 Nilsson, 2009 #72, 200-202, 204, 209, 211, 214-217, 219-221, 223-225]) and social skills (n = 16 [168, 169, 173, 179, 186, 187, 192, 202, 206-208, 212, 214, 215, 222, 223]). Mental ill health, often referred to as psychiatric problems, was an exclusion criteria in nine studies.[191, 194, 198, 201, 209, 210, 215, 224, 225]

The delivery of interventions occurred across a variety of settings and by a range of personnel (See Supplementary Materials 2, Table 5). Twenty of the 57 studies reported on interventions delivered in a hospital or clinical setting [118, 180, 181, 187, 191, 193, 194, 196, 199, 201, 203, 205, 206, 208, 209, 211-213, 215]. Other frequent settings were online (13 studies [175, 182, 188, 197, 198, 202, 216, 217, 219, 221, 223-225]) and campsites (ten studies [168, 170-172, 174, 178, 179, 184, 190, 220]), this included one psycho-education intervention[220] delivered during a residential camp. Interventions were delivered by mentors, various clinicians, music therapists, researchers, counsellors, volunteers, students, social workers, the recipient themselves, parents and a martial arts instructor. Twenty-six studies involved interventions delivered at least in part by clinicians with specialised training [169:Brothers, 2014 #1656, 175, 177, 179, 181, 183, 184, 186, 191, 192:Curle, 2005#1662, 194, 200, 201, 203, 205, 207-211, 214, 217, 219, 220]. In ten studies interventions were multi-disciplinary, delivered by more than one specialist[169, 178-180, 186, 191-194, 220]. Although not widely reported, 16 studies reported that practitioners delivering the intervention received supervision or training.[167, 169, 171, 179, 180, 182, 183 , 190, 191, 193-195, 200, 206, 208, 214, 218]

Where the structure of interventions was reported (n = 48), this indicated that the length of interventions was often at least two months, commonly with weekly or twice weekly sessions. Where interventions occurred once, they were typically camps that often lasted for around a week or procedural interventions that after introductory session/s lasted for the length of the procedure. A number of interventions were ongoing, that continued beyond the study in question.[169, 177, 180-182, 188, 193, 194, 196, 200, 201, 203, 206-210, 212, 215-218, 221, 225]

Quality appraisal

A summary of results of the quality appraisal of included articles are displayed in Table 4. Quality ratings were based upon individual articles rather than studies to allow for the different research questions across articles. Only one of the 60 articles, a PhD dissertation, scored a "Yes" against every quality appraisal question.[169] Thirteen articles scored a "yes" for at least 12 quality appraisal questions.[169, 171, 174, 183, 189, 193, 200, 205-207, 209, 214, 222] Whittemore and colleagues scored poorest with nine "no" ratings indicating rather lower study quality than other articles.[202] The Stewart[175] article reported on the same study as Barnfather,[214] the latter article scored well on quality appraisal with only a "no" for consideration of limitations of methods and a "cannot tell" rating for rigorous data collection. This discrepancy in quality appraisals illustrates the difficulty in rating quality, where different journals, authors and disciplines may have different criteria for reporting. The Bignall article is an RCT included in Review 1.[118]

With regard to the criteria against which articles were appraised, articles were most likely to score positively in relation to findings being substantiated by data, clarity of research question, study design appropriateness, adequacy of sample and description of data collection. . We were not able to determine whether the authors' theoretical perspective had influenced the study design, methods or research findings in 45 articles , suggesting a lack of reflexivity in the reporting of the included studies. Twenty-nine articles were at risk of bias in relation to the context or setting of qualitative research being inadequately described.[118, 167, 170, 172, 173, 175, 178, 179, 181, 182, 184, 186, 188, 189, 195-198, 201-203, 208, 212, 215, 217-220, 223] Twenty-two articles were rated as being poor quality regarding the clarity of description of interventions.[170, 172, 173, 175, 176, 181, 187, 188, 192, 204, 206, 208-213, 217, 218, 221, 223, 225] After excluding theoretical or ideological perspective, which was rarely reported by included articles, five articles were of excellent quality scoring a "yes" or "cannot tell" for all other quality appraisal items.[169, 171, 200, 205, 222]

Table 4: Quality appraisal of included qualitative articles

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Ayers et al., 2011	Y	N	?[a]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
Barlow et al., 1999	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	N
Barnetz et al., 2012	Y	N	?	Y	N	Y	Y	N	Y	Y	Y	Y	?	Y
Barnfather et al., 2011	Y	Y	Y	Y	Y	Y	Y	?	Y	Y	N	Y	Y	Y
Barry et al., 2010	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Baruch, 2010	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	N
Bignall et al., 2015	Y	N	?	?	N	Y	N	?	N	Y	Y	N	N	Y
Bluebond-Langer et al., 1991	N	N	?	Y	Y	Y	Y	?	N	Y	N	Y	Y	Y
Brodeur, 2005	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Brothers et al., 2014	Y	N	?	Y	N	Y	N	Y	N	Y	Y	N	N	Y
Bultas et al., 2015	Y	N	?	Y	N	Y	Y	?	Y	Y	Y	N	Y	N
Burns et al., 2010	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Campbell et al., 2010	Y	N	?	Y	Y	N	Y	Y	Y	Y	Y	Y	N	N
Curle et al., 2005	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Y	Y
Dennison et al., 2010	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
Desai et al., 2014	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Docherty et al., 2013	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	N	Y	N
Fair et al., 2012	N	N	?	Y	Y	Y	Y	?	Y	Y	Y	Y	N	Y

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Gan et al., 2010	Y	N	?	Y	Y	Y	Y	?	N	Y	Y	Y	Y	Y
Gaysyn-sky et al., 2015	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y
Gillard et al., 2011	Y	N	?	Y	N	Y	Y	Y	Y	Y	N	Y	Y	Y
Gillard et al., 2013	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	N	Y
Gillard et al., 2016	Y	Y	Y	Y	N	Y	Y	N	Y	Y	Y	N	Y	N
Griffiths et al., 2015	Y	Y	Y	Y	N	Y	Y	?	Y	Y	Y	Y	Y	N
Hosek et al., 2012	Y	Y	Y	Y	N	Y	Y	Y	Y	Y	Y	N	N	N
Jaser et al., 2014	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	N	Y
Kashikar-Zuck et al., 2016	Y	N	?	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Kirk et al., 2016	Y	N	?	Y	N	Y	Y	Y	N	Y	N	Y	Y	N
Lewis et al., 2016	Y	N	?	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Mac-Donald et al., 2010	Y	N	?	Y	N	Y	N	Y	Y	Y	Y	N	Y	N
Marsac et al., 2012	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	N	Y
Masuda et al., 2013	Y	N	?	Y	N	Y	Y	?	N	Y	N	N	Y	Y
Moola et al., 2015	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Muskat et al., 2016	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	N
Nicholas et al., 2007	Y	N	?	N	N	Y	N	Y	Y	Y	N	N	?	Y
Nicholas et al., 2009	N	N	?	Y	N	Y	N	?	Y	N	Y	?	Y	Y

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Nicholas et al., 2012	N	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	Y
Nieto et al., 2015	Y	N	?	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	Y
Nilsson et al., 2009	Y	N	?	Y	Y	Y	Y	?	Y	Y	N	Y	Y	Y
O' Callaghan et al., 2011	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
O' Callaghan et al., 2012	N	N	?	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	N
O' Callaghan et al., 2013	Y	N	?	Y	Y	N	Y	Y	Y	Y	Y	N	Y	N
Reme et al., 2013	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Romero, 2014	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
Serlachius et al., 2012	N	Y	Y	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	Y
Shrimpton et al., 2013	Y	N	?	Y	N	N	Y	Y	Y	Y	Y	Y	Y	N
Sibinga et al., 2011	Y	N	?	Y	N	N	Y	Y	N	N	Y	Y	Y	Y
Stewart et al., 2013b	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
Stewart et al., 2013a	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Stewart et al., 2011a	Y	N	?	Y	N	Y	Y	Y	Y	Y	N	N	Y	N
Stinson et al., 2010	Y	N	?	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Y
Stinson et al., 2008	Y	N	?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N

	Is the research question clear?	Is the theoretical or ideological perspective of the author (or funder) explicit?	Has this influenced the study design, methods or research findings?	Is the study design appropriate to answer the question?	Is the context or setting adequately described?	Is the sample adequate to explore the range of subjects and settings, and has it been drawn from an appropriate population?	Was the data collection adequately described?	Was data collection rigorously conducted to ensure confidence in the findings?	Was there evidence that the data analysis was rigorously conducted to ensure confidence in the findings?	Are the findings substantiated by the data?	Has consideration been given to any limitations of the methods or data that may have affected the results?	Do any claims to generalisability follow logically and theoretically from the data?	Have ethical issues been addressed and confidentiality respected?	Are the interventions of interest clearly described?
Tiemens et al., 2007	Y	N	?	Y	N	Y	N	Y	Y	Y	Y	Y	N	Y
Weekes et al., 1993	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	N	N	N
White et al., 2016	Y	N	?	N	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
White, 2014	N	Y	Y	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y
Whitte-more et al., 2010	Y	N	?	Y	N	N	N	N	N	Y	N	N	N	Y
Wolf Bordonaro, 2005	Y	Y	Y	N	N	N	Y	?	N	N	Y	N	Y	Y
Wright et al., 2004	Y	N	?	Y	N	N	Y	Y	Y	Y	Y	N	Y	Y

Y = yes; N = no; ? = unclear; green cell denotes positive score; yellow cell denotes unclear or moderate score; red cell denotes negative score.

Qualitative synthesis

The translation of first and second order construct data across all sixty articles yielded five third order constructs; 'A Therapeutic Foundation', 'Social Support', 'A Hopeful Alternative', 'Resilience' and 'Getting In and Staying In'. A number of third order themes contribute to each construct. The line-of-argument that suggests how the constructs and themes are related to one another is represented pictorially in . This line-of-argument is described in detail at the end of the qualitative synthesis. Important areas of overlap between themes are illustrated by intersecting circles. Arrows between constructs and themes suggest how they may relate to one another.

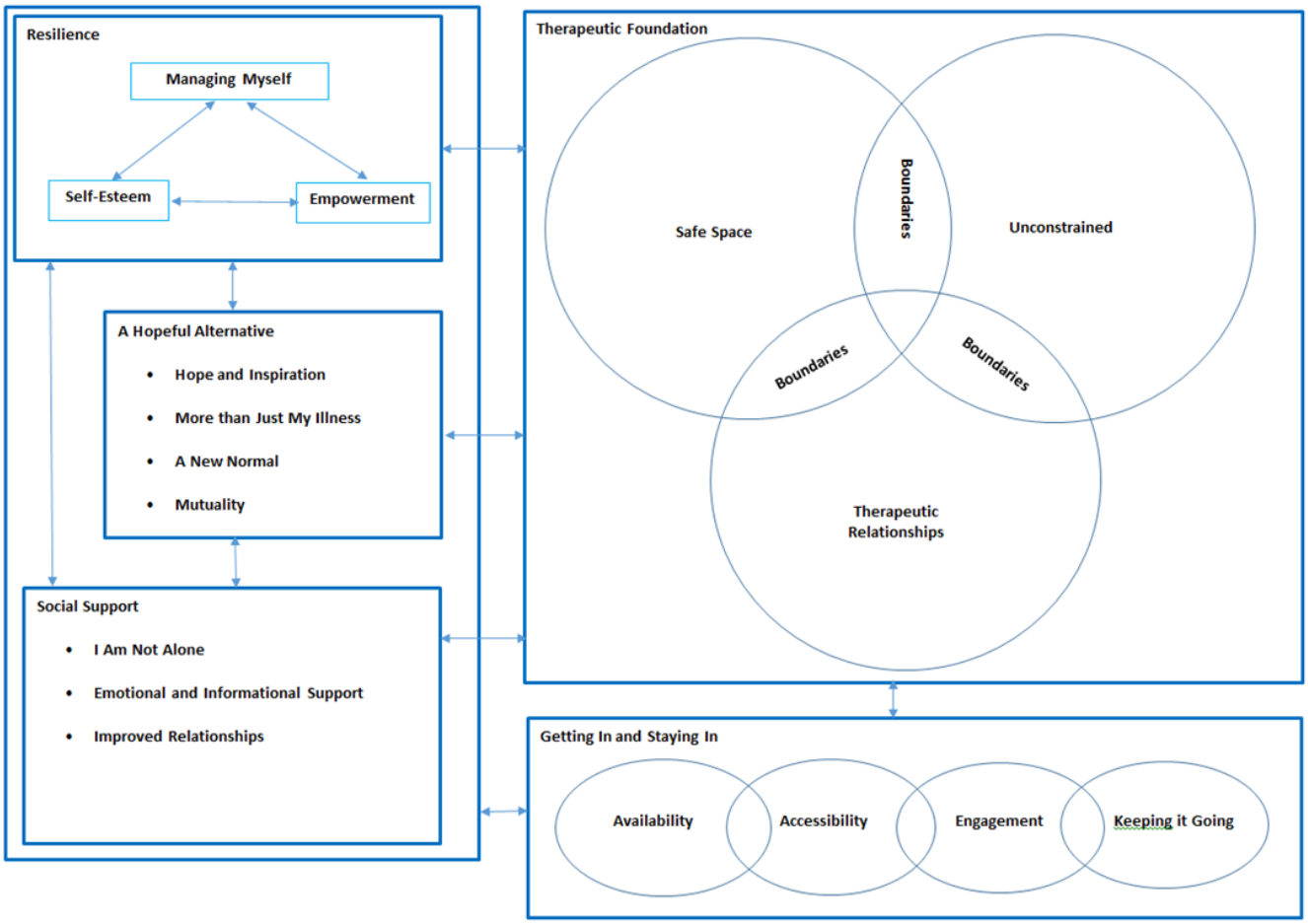

Figure 18: Line-of-argument synthesis

Supplementary Materials 2, Table 6 provides an overview of the themes within each construct and the articles which contribute to each theme. The author interpretations or second-

order ideas that contributed towards each third-order theme are discussed within each section, with first-order quotes in italics presented for illustration purposes and to maintain the link with participants' voices. Each quote is accompanied by a label within square brackets, acknowledging the type of participant providing the quote and the LTC under consideration within the study the quote was taken from e.g. [Parent, Juvenile Idiopathic Arthritis]. Additional interpretation is provided where necessary after first-order construct data, to explore its contribution to the theme.

Further detail regarding the third-order ideas which contribute towards each theme and the first and second-order data these ideas are based upon can be found in *Supplementary Materials 2, Table 7*.

Construct: A Therapeutic Foundation

A 'Therapeutic Foundation' describes how mental health interventions can confer a sense of safety for CYP, through the provision of an environment which is perceived as physically and emotionally secure. The content of the four themes within this construct explores a range of factors which may contribute towards the development of a therapeutic space. The first theme 'A Safe Space' discusses the importance for CYP of knowing what to expect and/or prior experience of an environment as safe. In order to perceive a space as safe, CYP need to know what is expected of them and have their privacy respected; needs which are explored within the second theme 'Boundaries'. The third theme 'Unconstrained' explores how CYP value the opportunity to escape from the worries and everyday routine associated with their LTC. 'Therapeutic relationships' is the fourth theme and explores different aspects of the relationship between CYP and their peers or practitioners which may be considered therapeutic. Overall, the 'Therapeutic Foundation' construct aims to emphasise the importance of CYP having access to somewhere where they feel able to express themselves fully and be listened to and understood by empathetic people.

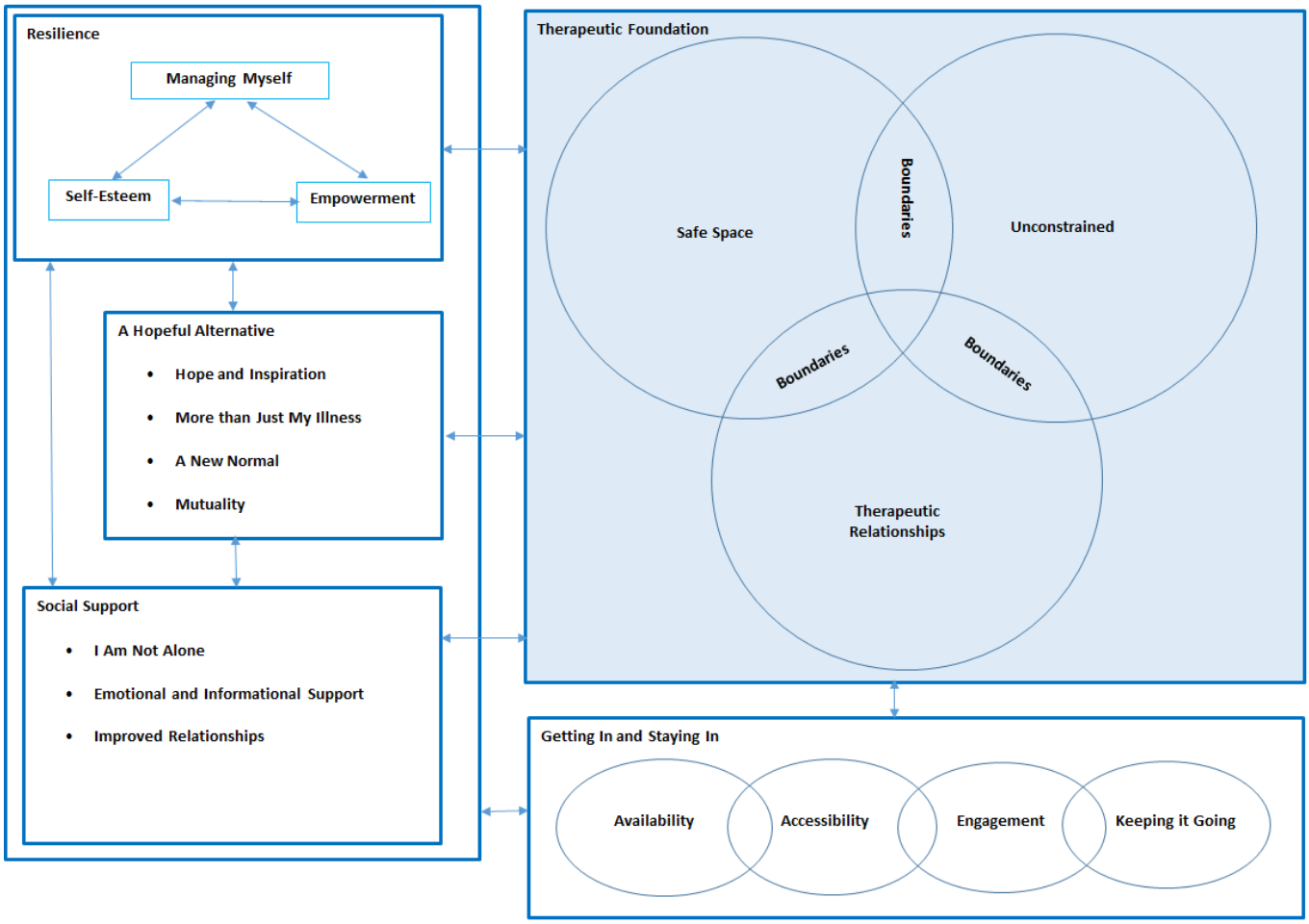

Figure 19: Inter-relationship between Safe-Space, Unconstrained, Therapeutic Relationships and Boundaries in creating a Therapeutic Foundation and links with other constructs

Figure 19 indicates how some second-order construct data contribute towards more than one theme. This overlap between 'Safe-Space', 'Unconstrained', 'Boundaries' and 'Therapeutic Relationships' aims to illustrate how the experience of one of these aspects may facilitate the occurrence, or experience, of another.

Theme: A Safe Space

'A Safe Space' illustrates how the presence of familiar family or staff, within a setting which is already known to them, can result in CYP feeling reassured and comforted. These feelings are necessary if CYP are to feel receptive to, and thus benefit from, an intervention. The people who deliver an intervention, or 'Intervention deliverers', have an important role in helping CYP familiarise themselves with the intervention setting and with other people taking part. . These factors, together with an understanding of what the intervention entails, may help

reduce anxiety about entering an unfamiliar environment and enable CYP to experience the setting as somewhere which is both emotionally and physically 'safe'. Nineteen articles contributed to this theme (See *Supplementary Materials 2, Table 6*).

Authors of three studies found that CYP may experience anxiety at the prospect of meeting unknown people prior to an intervention[177, 182, 187] and that the presence of a parent or support of a known staff member, such as a social worker, can reduce this anxiety and enable CYP to engage with a previously unknown intervention (see *Supplementary Materials 2, Table 7*).[171, 177, 187] A CYP with T1DM illustrates this when discussing their anxiety about meeting the person delivering their psycho-education intervention for the first time:

> *... I hadn't met anybody and didn't know where I was going and all those kinds of things, ummm, being quite young, being very ill, very worried. And I personally wouldn't have coped very well if they had just invited me to go to somewhere. I needed my mum there* [Participant type: CYP, LTC: T1DM] [187](p.175)

The above suggests that the involvement of someone who is known to the CYP appears to be comforting. We suggest that the reduction in anxiety as a result of the presence of someone familiar may potentially 'mask' a mental health need of CYP taking part. However, within the initial stages of an intervention, a familiar person could transform a previously feared space into one that is 'safer', allowing CYP to better engage with the intervention being delivered. The following quote from a CYP taking part in a study examining different management techniques for needle related distress illustrates this:

> *If I actually knew the nurse it would be okay, but if I didn't I would be like 'what's going on? I don't even know you!'* [CYP, Cystic Fibrosis] [204](p. 337)

We suggest that making an environment feel safer may be an important intervention itself, especially for CYP with anxiety around medical procedures. This may depend on the frequency and duration of medical treatments and related levels of distress for the CYP.

Fears about not knowing anyone else who was taking part were also acknowledged at the start of a creative writing group,[177] and online peer-support interventions.[182] Authors of three studies suggest that CYP valued the opportunity to get to know peers more.[167, 179, 184] As CYP became more acquainted with one another a feeling of acceptance was created, giving the

intervention space a sense of being familiar and safe, helping CYP feel able to share their feelings and experiences with one another. This is illustrated by a camp participant below:

> *The camp did help with that [to talk about our different experiences] by bringing us all together and letting us get to know each other for a couple of days so that we felt that we were comfortable enough to go up to each other and just talk about it.* [CYP, Craniofacial differences] [184](p. 64)

First and second-order data from eleven studies suggests that the individuals who delivered the interventions played a key role in supporting the initial recognition of similarity between peers and encouraging interaction and engagement during the early stages of group interventions.[171, 177-182, 184, 189, 197, 202] Second-order data from five studies suggests deliverers of interventions should plan to allow sufficient time for CYP to get to know each other.[171, 174, 179, 180, 184]

Supplementary Materials 2 indicates that prior experience of a setting as somewhere safe was viewed positively by CYP.[180, 181] Below a CYP indicates how they perceived coming back to a hospital environment with which they were already familiar for an introduction to a peer support programme:

> *…for me, the hospital became like a second home. It was my safe haven. I always felt safe in the hospital. I always felt comforted in the hospital. So having the 8-week group in the hospital, I saw it as somewhere safe that I could come back to.* [CYP, Mixed] [180](p. 2549)

Authors identified that CYP valued knowing what to expect from an intervention, either through explanation regarding the content of the intervention and its aims by the intervention deliverer, [187, 190, 200] or by providing the opportunity to observe others participating before taking part themselves.[211] Wolf-Bordonaro[203] discusses how the process of asking for information from the intervention deliverer about the time and place of follow-up sessions by CYP may be a method used by CYP to feel in control within the intervention situation. Many CYP with LTCs experience unpredictable symptom fluctuations and are expected to attend appointments to manage their physical health; aspects of their life over which they may have little control. We suggest that feeling in control of a situation related to their LTC could be a novel experience for CYP, providing an opportunity to distance themselves from anxiety

associated with their LTC and its management. CYP may feel reassured by the presence of a predictable intervention structure, as illustrated by Gillard and colleagues [179] who discuss the use of ground rules and set format within their "TeenTalk" intervention. This use of predictability to help CYP feel safe within the intervention setting is explored further within 'Boundaries' below.

This theme indicates that CYP may find it helpful to invite their family members and familiar staff to attend distressing procedures, and accompany them in unfamiliar intervention situations. It is also important that CYP are given enough time to get to know one another in group settings and are supported to familiarise themselves with the setting and staff beforehand. Knowing what to expect from an intervention through use of a predictable routine and requesting information may also help reduce CYP's worries about attendance and help them feel safe and in control of the intervention situation.

Theme: Boundaries

This section considers the theme 'Boundaries.' Authors of the 34 articles included in this theme (see *Supplementary Materials 2, Table 6*) describe the efforts made by the deliverers and/or recipients of an intervention to set certain rules for one another. These boundaries may include; maintaining the privacy and anonymity of intervention recipients, setting rules for behaviour or acceptable discussion topics and having an established meeting time, structure and setting. As indicated within *Figure 19*, the setting of boundaries appears to facilitate the experience of a setting as 'safe', which may in turn enable CYP and their families to discuss their emotions and sensitive issues connected to living with their LTC. This is an example of CYP feeling 'unconstrained' by their LTC, an idea which is explored in the following theme.

Seven studies suggest that among CYP with LTCs there appears to be a fear of their diagnosis, or other issues they regard as private, being disclosed to others.[172, 177, 179, 182, 183, 192, 201] Authors describe health and social care professionals playing an important role in advertising interventions to an appropriate audience,[177] monitoring those who are able to access the intervention [216] and locating a secure, safe environment in which to discuss sensitive topics.[181]

Three study authors suggest there is a demand for intervention deliverers to respect the privacy of CYP by creating opportunities for CYP to talk without parents or the deliverer present,[184, 187, 208] as recognised by a parent supporting their child to attend a CBT intervention:

> *I did wonder whether they should have spoken to her by herself. […] I just felt like there could be things going on that are of worry and whether friends like you etcetera that may not want to be said in front of parents* [Parent, T1DM] [187](p. 175)

This appears to contradict the observation made under 'safe space' that the presence of a familiar person may enable CYP to access an intervention and talk about their experiences. We suggest that as CYP become familiar with an intervention and its setting, they become more open to meeting with an intervention deliverer, or other young people, without the support of family or known staff members.

Two studies suggested that online interventions provide the opportunity for greater anonymity than face to face interventions, which some CYP and their parents felt facilitated participation and engagement among less confident attendees.[182, 183] This is evidenced by a quote from a CYP who took part in an online support and education, supported by older peer-mentors:

> *The Internet was better because you're not looking at them straight in the eye. I guess it is a bit easier. I think I shared my feelings better. Face-to-face would be hard.* [CYP, Asthma/Allergies] [183] p. 188)

Three studies suggest that the use of certain rules, such as use of a deliberate turn-taking structure within sessions, can help CYP feel safe enough to express their emotions, [179, 194] as well as ensure that these emotions were appropriately managed.[192] This is illustrated by a family member discussing taking part in a family-system intervention:

> *I liked how each of us had a turn to speak and share our experiences. I also liked how everybody else listens on what you have to say* [Sibling, ABI] [194](p. 659)

Whilst Gan and colleagues[194] do not indicate how the precedence for this turn-taking was established, the topic manual developed to guide the implementation of their family intervention indicates that time to introduce the intervention and develop "an agreed contract

between participants and intervention deliverers during the early stages of the intervention. One mother taking part in a family art-therapy intervention agreed that boundaries around acceptable behaviour should be established early on in the intervention:

> *The kids, you know, I think it's extremely important when you are dealing with children, regardless of illness or age, that they know from the get go, what is expected, and what their limitations are[…]then when they are imposed after the fact, it feels restrictive*. [Parent, Mixed] [169](p. 197)

Here the late introduction of boundaries to curb potentially disruptive behaviour appears to have been experienced as critical and restraining. Perhaps the introduction of boundaries at an earlier stage of an intervention may ensure CYP behaviour remain both respectful of others, but also enable them to relax, have fun and feel 'uncontrolled'. This possibility is supported by Gillard [178] who highlights how camp staff supported CYP to take part in "transgressive activity" (p. 895) whilst attending camp, such as the challenge to overcome "security" measures to win a box of sweets for their cabin group. This was seen as an opportunity to engage in risk-taking behaviour within set appropriate limits. Four studies highlighted how behavioural rules ensured that CYP respected the personal space and boundaries of others,[178, 186, 190] which contributed towards the creation of a supportive community within peer group interventions:

> *That it's got a sense of community…that everybody respects everybody…and you can talk about everything and anything…Some people disagree with you, but they don't bark at you.* [CYP, CP/SB] [175](p. 798).

One topic where discussion appeared to be restricted was in relation to life-threatening LTCs. Within some interventions, the avoidance of the discussion of death and dying was noticeable. For one study, this was a result of the interventions explicit aim to provide the child with respite from worries regarding their LTC,[178] whilst within an online support site for CYP with CF and their parents, the avoidance of discussing the life-limiting nature of the LTC was not explicitly discussed. [188] Perhaps this reflects the desires of participants within this group and may highlight the need to discuss ground-rules at the beginning of an intervention, to ensure participants know what to expect.

The conscious or unconscious avoidance of discussions of death may reflect the need of CYP with certain diagnoses, and those involved with their care, to change the focus of mental

health and wellbeing interventions to consider 'how to make the best' of the time left and live as normal a life as possible. In contrast, two studies aimed at alleviating distress for CYP with cancer undergoing stem-cell transplantation suggest the interventions provided the opportunity to confront their own mortality and the possibility of dying.[207, 208] These two apparently contradictory concepts illustrate the important role mental health interventions may play in facilitating both the acceptance of the possibility of death and ensuring life is as fulfilling as possible for CYP with potentially terminal diagnoses. We suggest that the balance between these two alternative outcomes may be affected by LTC type and prognosis, and the choice of coping strategy preferred by the CYP and their family. The evocative nature of the work, particularly in supporting CYP at the end-stage of their illness, necessitates the presence of an intervention deliverer who is adequately trained and the resources to ensure that they are provided with adequate supervision. The need for an intervention to be adequately resourced and consider the type of LTC and personal preferences of CYP is discussed within the '*Availability'* and '*Engagement'* themes under '*Getting In and Staying In*'.

Setting aside time to spend together can be difficult due to the challenges of meeting the needs of different family members and balancing family life alongside the demands of managing an LTC.[169, 193] First and second-order construct data from two studies evaluating family-orientated interventions indicated that CYP and their parents appreciated having a scheduled opportunity to spend time together as a family. [169, 194] This appeared to facilitate communication between family members, as described by two parents taking part in a family system intervention, facilitated by a social worker/rehabilitation counsellor:

> *Opened my eyes as to what [the adolescent with the ABI] was thinking* [Parent 1, ABI]
>
> *Helped us to discuss concerns and feelings that otherwise might not surface* [Parent 2, ABI] [194](p. 659).

This opportunity to communicate more openly facilitated insight into what the CYP with the LTC was thinking and meant the family could use the space to better understand each other's feelings. Authors of two studies also indicated CYP valued the opportunity to meet with peers.[168, 179] This may prove beneficial for CYP in building respectful, understanding and

empathetic relationships with family and peers with an LTC, as discussed below under '*Social Support*'.

Within this section we discuss the value of establishing boundaries, and their role in ensuring that CYP experience an intervention setting as safe and feel able to express their emotions. Intervention deliverers and participants can be involved in the creation of boundaries, which through the sharing of experiences can facilitate the development of understanding and caring relationships.

Theme: Unconstrained

This theme explores how the opportunity to meet similar peers seemed to be important to CYP. This experience appeared to provide CYP with the freedom to fully express their emotions without being constrained by the expectations or views of others. The theme also discusses how mental health and wellbeing interventions can provide an experience of freedom or respite from the constraints and daily routine of having an LTC and the anxiety and distress which can result. Twenty-eight articles contributed to this theme (See *Supplementary Materials 2, Table 6*).

Whilst the idea of an intervention setting being both 'safe' and 'unconstrained' appears to be contradictory, there is a large degree of overlap in the 12 studies that contribute towards both of these categories,[169, 171, 178-182, 184, 186, 211] across a diverse range of interventions and LTCs. [167, 187] The 'Boundaries' category suggests that a therapeutic space must have limits if it is to be experienced as safe by CYP. This may account for these two different sets of ideas from the same studies. The use of 'boundaries' within an intervention may need to be flexible to consider the changing needs of participants and facilitate their ability to access 'A Safe Space' or feel 'Unconstrained' as required. A possible example of this is that CYP may wish to think about the risk of dying on some occasions, but not others.

Interventions which provided the opportunity for CYP to be themselves, speak openly and experience freedom from stigma or judgement were particularly valued[168, 171, 172, 174, 178, 179, 181, 192] and facilitate a safe space where CYP felt able to talk and share their experiences:

> *I feel open that I could talk to whoever, however I want like I can talk open about what I feel and what I need.* [CYP, HIV] [181](p. 6)

One way in which this appears to have been achieved is by CYP interacting face-to-face with peers with similar LTCs (See *Supplementary Materials 2, Table 7*). Six of these interventions were residential camps, [168, 171, 172, 174, 178, 179] however this opportunity was also provided within two weekly peer-support group interventions.[181, 192]

The opportunity to feel free from the perceived judgement of others appeared to be particularly relevant for CYP whose LTCs result in a visible difference in their appearance, or which are associated with high levels of perceived stigma.[168, 171, 172, 177-179, 181, 192] Whilst the majority of the interventions included in this review were delivered to CYP with the same LTC, there was some evidence to suggest that CYP, including those with stigmatised diagnoses,[172] also enjoyed and benefited from interacting with peers with different LTCs.[169, 182, 183, 223] This evidence suggests that the ability of CYP to empathise with others who are experiencing similar difficulties to them may transcend diagnosis.

In addition to the positive benefits of spending time with peers in similar situations, authors of five studies explicitly discussed the value of CYP having the opportunity to access an 'adult free space' to meet by themselves[174, 184] or separately with the intervention deliverers. [169, 187, 208] Four of these studies interviewed participants between nine and twenty-four years of age, thus the developmental stage of participants may have influenced their expressed need and comfort with a space separate from their parents. [174, 184, 187, 208] A flexible approach, depending on timing and needs of CYP might be appropriate, as suggested by CYP with craniofacial conditions receiving a camp intervention:

> *It is good to have a leader [adult deliverer] but maybe not [one] who does everything . . . like especially when we didn't know each other at first it was really tough to get us to talk...So it might be good to have a leader at first but once we got to know each other it was a little easier.* [CYP, Craniofacial conditions] [184](p. 67)

These peer relationships also appeared to provide CYP with a release from perceived pressure from health professionals and parents, as illustrated by a CYP with T1DM talking about their relationship with their mentor, a slightly older peer with lived experienced of their LTC.

The mentor manages to come into both worlds, the world of the adults and the world of my worries, he knows them, you can choose what to share with him and what not to share, he's tolerant. He doesn't pressure me. [CYP, T1DM] [167]p475

In contrast with the need for structure highlighted within 'Boundaries', Barnetz and colleagues highlight how young adolescents can learn to accept their condition through relationships based upon enjoyment and interest, unconstrained by the adult-orientated approach to relationships which emphasises structure, contribution, and development.[167] This apparent freedom from adult rules offers a contrast to life as usual for CYP; where opportunities for experiencing autonomy may be complicated or limited by the LTC. The ability to learn and express feelings through unstructured, informal contact with peers was also identified by studies evaluating camp[168, 171, 174, 179, 184, 190] and technology based interventions.[182, 188] The role of CYPs being able to express themselves emotionally in order to receive emotional support and learn new skills is discussed in more detail within the '*Informational and Emotional Support*' and '*Managing Myself*' themes below.

Another way CYP were able to feel "unconstrained" by their LTC was through interventions which provided respite from the "daily suffering and uncertainty" associated with treatment for an LTC, [208](p. 6), as well as a temporary reprieve from worries about daily life and the limitations enforced upon them by the LTC. This is illustrated below by a quote from a parent of a child receiving music therapy:

He looked forward to the [therapeutic music session]. He knew it was going to be a day where he didn't have to worry about if they were coming in to give him a pill or if they're coming in to start the chemotherapy [Parent, Cancer] [207](p. e24).

Interventions associated with feelings of respite seem to be those that are not part of routine treatment, such as camps, or interventions which aim to provide an alternative environment or distraction from the hospital setting.[182, 207, 211] This release from the daily burden of living with an LTC could potentially improve the mental health of CYP. CYP taking part in camp interventions spoke of the bodily freedom they experienced in being able to take part in activities that they had never tried before, due to the physical limitations placed upon them by their LTC and risk management strategies used by adults they came into contact with.[170, 171, 174, 178, 179] Authors of five camp studies discussed how, due to adaptations such as

the provision of trained staff and specialist equipment to support physical health needs, [170, 171, 178, 179, 190] CYP were able to take part in an activity commonly participated in by American CYP without LTCs. This brief opportunity to experience being like any other child may be a novel experience for CYP with LTCs and is discussed further under '*A New Normal*' below.

The emphasis on how much CYP enjoy fun and engaging activities was acknowledged by 18 studies[167-173, 177-179, 182, 183, 190, 192, 194, 202, 211, 212] and could be considered as another important factor which allows CYP to relax and "just be a kid" [178](p. 897). This is an important component to consider when designing interventions which CYP find engaging, discussed later within the '*Engagement*' theme. However, it may not always be appropriate to incorporate an element of 'fun' into a mental health intervention. An example of this may be for CYP receiving procedural interventions, where the focus of a mental health intervention may be on reducing the distress of CYP enough to support them to engage with the procedure. In addition, three study authors acknowledged that there was the risk of CYP learning information about their illness or those of others which may upset them whilst interacting with peers with similar LTCs.[168, 182, 190] For example, one CYP with cancer talks about the uncertainty created through meeting peers who had experienced a relapse in their illness at camp:

> *It worries me sometimes, because they [campers who relapsed] thought that someday they'd be sure, and they could go on with their lives. And it didn't turn out that way. So once in a while, I think, "Well, maybe I will relapse, and I won't be around very long."* [CYP, Cancer] [168](p. 77)

Nicholas and colleagues [182] highlight that having a member of staff to address any anxiety or distress that may be created through peer-interaction was important.

The studies contributing to this theme indicated that CYP appreciated the chance to speak openly, particularly when they did not often have this opportunity within their everyday lives.[172, 179] CYP also valued opportunities to escape the routine and pain associated with their LTC through new environments, having fun and trying new activities.

Theme: Therapeutic Relationships

This theme, receiving contributions from 42 articles (see *Supplementary Materials 2, Table 6*), characterises therapeutic relationships as the availability of trustworthy others, who

make CYP feel cared for, respected and valued. These relationships offer CYP the opportunity to share their stories and be listened to in a respectful environment. Studies included in this review indicate that relationships of this quality are not just limited to interactions between CYP and a trained intervention deliverer. This theme also considers the importance of relationships with peers and families in similar situations in helping them to feel secure within the intervention setting.

Authors of seven studies note that knowing helpful people were consistently available was valued by CYP receiving interventions[167, 171, 179, 180, 188, 191, 203] (See *Supplementary Materials* 2). This is illustrated by a CYP describing their perception of counsellors at their camp:

> *That is what camp means to me, I love the counsellors. They are awesome and if you need help just ask them a question and they will help you.* [CYP, CHD] [171](p. 557).

Within the camps included in this review, camp counsellors referred to volunteers, often previous camp attendees,[172, 178] who provided support and supervised CYP whilst they completed activities. This suggests that therapeutic relationships can exist between CYP and individuals without specialised mental health training.

Along with interest and understanding,[191, 195, 201,216, 225] *Supplementary Materials 2, Table 7* indicates that mental health and wellbeing interventions provided the opportunity to build empathetic connections with both peers and intervention delivers.[188, 202, 211] This opportunity is evidenced by a CYP discussing an internet-based coping skills programme with a discussion board component:

> *The scene presented was true…but, most of all I liked knowing that other teenagers know how I feel. That made me feel good.* [CYP, T1DM] [202](p. 8)

These empathetic interactions appeared to provide the opportunity for CYP to receive recognition and validation of their experiences.[168, 169, 177, 180, 182-184, 191] First and second-order construct data from ten studies suggest that these connections may elicit feelings of being cared for and valued.[169, 172, 174, 177-179, 181, 183, 190, 203] This is evidenced by a CYP describing the emotional effect of their relationship with fellow campers:

I would probably take that feeling of like being wanted because I know, like, for some kids […]they don't feel as, like, wanted or like people want to be around them. […] it's not, like, very often that you're included in a lot of stuff. [CYP, Mixed] [172](p. 116)

This suggests that it is important that CYP feel that they matter to other people who can see and value them for the person they are, beyond their LTC. This may help CYP build trusting relationships with their peers and intervention deliverers, to express themselves and thus create the opportunity for them to receive social support from peers as discussed under the *Social Support* construct.

This opportunity for self-expression and being heard by an empathetic audience, may in turn have reinforced the experience of the intervention space as safe and trustworthy. For example, a parent observed the following of the relationship between their child and the deliverer of a movie making programme designed to reduce CYP's anxiety whilst receiving radiotherapy:

She placed a lot of trust in them and she really loved them … And I think because of that she felt safe. She felt she was not going to be let down and that she could go through the process with the people around her. [Parent, Cancer] [212](p. 3).

These therapeutic relationships, including the positive regard of both peers and those delivering the intervention,[203] influence both how the child feels about themselves and their coping outside of the intervention setting. This is illustrated by a CYP discussing how their camp experience could affect them during their everyday life:

The love that I received here made me a better person because I was able to have more independence yet still have a lot of fun whenever I want and always find the bright side of things instead of things that are negative and pessimistic.[…], have more confidence in myself [CYP, Mixed] [172](p. 117).

The evidence within this theme suggests that relationships with empathetic peers, intervention deliverers and family members can play an important role in helping CYP feel validated, cared for and valued. This appears to facilitate the expression of emotions and increase the likelihood that CYP's psychological needs will be met. As a result, high-quality therapeutic relationships could be considered as a critical component of mental health interventions in their own right.

Summary: Therapeutic Foundation

The importance of a sense of familiarity, feeling unconstrained, establishing boundaries and developing therapeutic relationships has been discussed in the context of creating a "therapeutic foundation". The creation of such a foundation is something which is needed to form the basis of an intervention. It provides a healing space where CYP can be themselves; reinforces the engagement of CYP with an intervention and enables them to access social support from their peers and intervention deliverers. In turn this social support, as discussed within the next theme, may reinforce that it is safe for CYP to talk about their feelings and experiences. Having a safe space and supportive peer network can also enable CYP to learn new skills and fresh perspectives to help them manage their LTC, which can contribute towards the building of resilience and hope, concepts which are explored within the overarching themes below.

Construct: Social Support

Three themes contribute to the construct of 'Social Support': 'I Am Not Alone'; Emotional and 'Informational Support' and 'Improved Relationships'. 'I Am Not Alone' details how CYP can experience feelings of connectedness and belonging through contact with peers with an LTC. 'Emotional and Informational Support' discusses how CYP can gain access to emotional reassurance and receive information to help them to manage their relationships with peers and family through contact with empathetic, understanding peers and intervention deliverers. The final theme "Improved Relationships" discusses how an intervention may improve CYP's relationships with peers who do not share their illness experience and family members.

The concepts identified within the themes which contribute towards the 'Social Support' construct can influence CYPs experience of a 'Therapeutic Foundation' but can also potentially affect CYP's lives outside of the intervention setting. The emotional and informational support received as part of the intervention may enable CYP to communicate more freely with family and peers and have a positive impact on these relationships. These improved relationships may both reinforce the experience of an intervention setting as therapeutic and potentially extend this experience of a therapeutic, safe space beyond the intervention setting, into their daily lives in the form of social support. This may have important implications for maintaining engagement with the intervention and any effect beyond its end-point; as suggested within the

overarching construct 'Getting in and Staying In' which are described in detail in later sections. *Figure 20* depicts the relationship between the construct 'Social Support' and the other constructs.

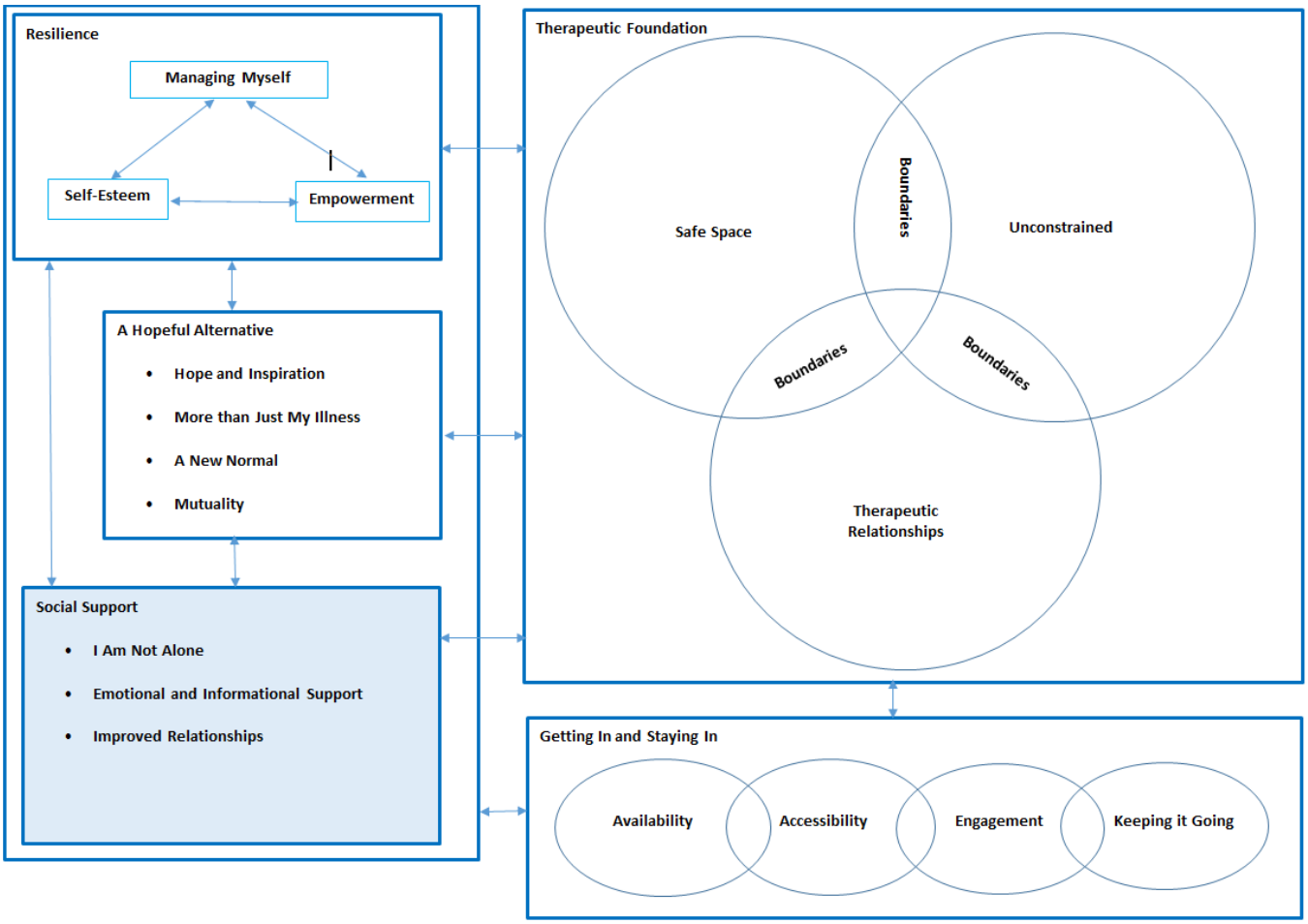

Figure 20: Therapeutic Foundation and Social Support

Theme: I Am Not Alone

This theme discusses how CYP value the opportunity to connect with others who are similar to them and the feelings of safety and belonging this can provide. Meeting and interacting with peers helped CYP to realise they were not the only ones living with such difficulties and normalised their experiences. This experience can help reassure CYP and reduce feelings of loneliness and isolation. Forty-six articles contributed towards this theme (see ***Error! Reference source not found.***, *Table 6*).

Six study authors discuss how, for some CYP, the experience of meeting people with similar experiences was a novel experience.[167, 171, 174, 177, 181, 190] Conditions such as CF actively

prevent CYP from meeting face-to-face in groups due to the potential risk to their physical health.[188] Other LTCs such as HIV and Sickle Cell Disease can be associated with high levels of stigma, poor understanding and grief within the wider-community, which can prevent CYP with these conditions from connecting with peers with the same conditions.[172, 181]

Ten studies indicated that CYP valued the sense of being understood and accepted through interactions with peers with similar conditions.[167, 168, 171, 176, 179-181, 184, 222, 223] This is illustrated by a quote from a CYP accessing a week long camp intervention:

> *At home I'm like Clark Kent…but like here, I'm Superman. I'm just like, I'm more open, more talkative, more inspired…'Cause when I'm at my home it's like I'm hiding from everybody else[…] I just don't communicate. Camp is more open, it's just friends all over.* [CYP, HIV] [179](p. 1514)

The quote suggests that feelings of being understood and accepted by similar peers may be related to CYPs perception they were free from stigma or judgement from others, facilitating feelings of safety and self-expression as discussed within '*Unconstrained*'.

Nineteen studies suggested that the opportunity to connect with others who share their experiences appears to facilitate feelings of belonging in CYP.[167, 169-171, 174, 176, 177, 179, 182, 184, 188, 191, 192, 194, 202, 206, 208, 222, 223] This is evidenced by experiences of a variety of interventions, that ranged from structured family art-therapy style or support groups for CYP[169, 177, 181, 194] to more unstructured interventions, such as camps:[171, 174, 179, 184]

> *here it's like a happy home so it's like, […] we are all like a family and our grandma lived here, and grandma invited us all over for dinner every three months, that's how it is, like whenever we come it's like 'Hey, good to see you.'* [CYP, HIV] [181](p. 9)

Authors of five studies indicate that the communication of experiences with peers or understanding intervention deliverers can help reduce feelings of isolation within CYP, [167, 171, 176, 191, 208] as supported by a quote from a CYP experiencing an online peer-mentoring intervention:

> *When I was doing the chats, I was involved with the conversation, and I wasn't thinking about [loneliness]…[The chats] made me realize that I'm not alone; like with my*

disability…there's lots of people out there that go through the exact same thing as I do [CYP, CP/SB] [175](p. 797)

These reduced feelings of isolation also occurred through use of online interventions,[182, 183, 188, 202, 222, 223] although some parents suggested the inclusion of an opportunity to meet either before or after commencement of online sessions would enhance bonding between CYP.[183, 222]

The reduced isolation through recognition that there were other people who were similar to them and the validation achieved by connecting through shared experiences could also be thought of as a form of "affirmational support" as conceptualised by Stewart and colleagues[183](p. 185). Stewart and colleagues describe affirmational support as CYP sharing challenges and solutions and thus receiving validation of their concerns and reassurance that they were not alone and able to cope well with their LTC.

Theme: Emotional and Informational Support

Forty articles contributed to this theme (see ***Error! Reference source not found.****2, Table 6*), which explores how both emotional and informational support are important components of social support experienced by CYP. We used Stewart and colleagues'[183] model of social support and their conceptualisation of "Emotional" and "Informational" Support to inform the set of ideas within this theme. In this section 'Emotional support' aims to encompass the relief and reassurance experienced by CYP as a result of expressing their feelings to peers who can empathise with them. Access to people with similar stories to tell and experienced intervention deliverers also enabled CYP to receive information and advice on how to manage and cope with their LTC. This informational and emotional support may reinforce CYP's experience of the intervention setting as a safe space where they can express their emotions freely and enable them to acquire skills and knowledge as discussed later within 'Resilience'.

Supplementary Materials 2, Table 7 indicates the opportunity to be in the same environment and share experiences with similar others enabled CYP to access emotional support and reassurance, [167, 177-179, 181-183, 188, 213, 222, 223] as illustrated by this quote from a participant attending a camping intervention for CYP with HIV:

You're just always getting reassured that it's okay that you have this [HIV], and it's just a good feeling. [CYP, HIV] [179](p. 1514)

CYP appeared to experience the sharing of their emotions with peers as a form of stress relief[179, 223] and valued the opportunity to talk about anxieties they worried that parents wouldn't understand. This is illustrated by a CYP with T1DM discussing their older peer-mentor:

> *You can share deep experiences and fears that other people won't understand or don't know how to calm.* [CYP, T1DM] [167](p. 475)

This suggests the presence of peers may facilitate emotional self-expression for CYP, due to their perception that their peers can understand what they have been going through, as discussed under '*Unconstrained'*. CYP may perceive that it is safe to discuss difficult feelings if they know others will be able to understand and empathize with them.

Authors of three studies suggest that those delivering interventions appear to have an important role in maintaining the 'emotional safety' of CYP during discussion of sensitive topics and reassuring CYP that other people will understand and listen to them.[183, 192, 194] Peers and intervention deliverers also provided CYP the opportunity to express and think about how to manage their emotions:

> *When I used to get angry, I just handle matters myself. I'd get in trouble. Here I don't get in trouble. They just talk to me for a while, about like, how it's not worth it.* [CYP, HIV] [179](p. 1518)

In addition to emotional support, CYP also expressed a desire for information on what to expect in the future, including information on; medications, activity limitations, variations in disease severity and possible impact on family, friend and romantic relationships.[173, 176, 194] This is indicated by a CYP discussing what they would like as part of a psycho-education programme:

> *I'd like to know what could happen, so you've got no surprises later on.* [CYP, JIA] [176](p. 603)

This suggests that informational support gained from individuals delivering an intervention may help reduce anxiety associated with not knowing what could happen next or how their LTC may impact on their lives in the future. The quote below from a CYP with HIV

illustrates how they intended to use the knowledge from a peer support group intervention to prepare them to answer questions from others in the future:

> *… …I always had to ask how do I get it again, how do I transmit it, […]and yeah cause I always had to ask the second questions because I always have to make sure that I'm at a point where if someone asked me, I'm ready to answer their questions.* [CYP, HIV] [181](p. 7)

Second-order construct data suggest that CYP valued information from same-age peers, older peer-mentors and intervention deliverers on how to manage relationship challenges such as teasing due to appearance or LTC management issues[171, 183, 222, 223] and how to manage issues of disclosure and sexual relationships.[173, 177] Listening to how peers managed these difficult situations may help inspire CYP through the recognition that they may be able to do this too, an idea explored under the theme '*Hope and Inspiration*'.

The data within this theme suggest that mental health interventions should aim to incorporate emotional and informational support components, as there is evidence that this may benefit CYP's mental health and wellbeing. These components may be delivered through contact with peers with similar conditions and/or intervention deliverers.

Theme: Improved Relationships with Family and Healthy Friends

This theme discusses how an intervention targeting CYP mental health may also improve connections with family members and healthy peers. This may occur through enabling CYP to help their family members and peers better understand their feelings and the impact of the LTC on their lives. Alternatively, interventions can directly target communication styles between CYP and family members or peers. Both of these approaches can improve relationships between CYP and key people in their lives. This may in turn result in increased social support for CYP outside of the intervention setting. Thirty-four articles contributed towards this theme (see ***Error! Reference source not found.****2, Table 6*).

Authors of four studies suggest that one method of changing dynamics within the family is by altering parenting styles, through both family directed interventions[169, 189, 194] and those focused on CYP.[190] Changes reported by parents as a result of these interventions included improved recognition of protective parenting-styles[190, 194] and increased attempts to focus on other aspects of family-life other than their child's LTC.[169]

Seven studies suggest that an intervention can provide a distraction from the LTC,[169, 182, 186, 195, 207, 211, 212] One CYP discussed how they enjoyed the alternative focus provided by a family art-therapy intervention:

> *It's a lot of fun. And I get to do things with my mom. And there are things, finally, that we can have fun at doing. When I was going through so much of my sickness, I wasn't able to do fun things. It's just a healthy experience.* [CYP, Mixed] [169](p. 171)

The idea that CYP appreciate interventions which consider other areas of their life in addition to their LTC is discussed further within '*More than just my Illness*'.

Interventions which aim to improve CYP mental health and wellbeing may also indirectly improve relationships between the CYP and other members of their family. Authors of 12 studies indicated that improved communication between family members and increased empathy for the child with an LTC resulted from interventions aimed at individual CYP, [189, 195, 201, 206-208, 211, 212] or peer groups.[177, 180, 182, 186] CYP receiving peer-support interventions valued advice from peers with similar conditions which helped them to resolve communication and relationship issues with family members [183, 222, 223] Our interpretation is that communication skills training may be an important component of a mental health intervention and be particularly relevant to young people transitioning into adulthood, with the resulting change in family dynamics.

The impact of interventions that directly and indirectly target family communication is illustrated by two quotes below. The first from a parent describing the impact attending a creative writing group for CYP with HIV had on her child; the second from a CYP discussing the impact a family intervention to support them with their ABI had on their family:

> *She opens up more to her mother and her family. She used to be pretty shy, but now she's like so talkative!* [Parent, HIV] [177](p. 386)

> *We as a family were able to come together to better understand my brain injury and how I live my life*, [CYP, ABI] [194](p. 659)

The above suggests that increased empathy and understanding of the impact the LTC had on the CYP and improved communication can improve family relationships.

Seven studies indicated how CYP and parents noted that relationships with healthy peers could also improve following an intervention:[168, 177, 183, 207, 208, 212, 223]

These improved relationships could stem from the creation of an age-appropriate means to tell their story and increase peer empathy (e.g. [206]) or through the acquisition of social skills learnt through communication with peers with LTCs (e.g. [183, 223])

Authors of nine studies discuss how the interventions evaluated helped CYP ask for the support they needed from family, peers and practitioners, or improved relationships with health professionals.[169, 182, 183, 196, 203, 206-208, 223] This is illustrated by a quote from a parent of a CYP taking part in an online intervention, discussing how the intervention influenced their child's relationship with their health-care provider outside of the online setting:

> *[through the online network the ill child] has gotten to know [a health care provider] a lot better so he feels much freer...to go ask her for help.* [Parent, Mixed] [182](p. 215)

As such, interventions which can support CYP to tell their story and engage in supportive interactions with peers and intervention deliverers may enable them to seek support when required from parents, healthy peers and health professionals.

There was some evidence *(See Supplementary Materials 2, Table 6)* to suggest that the impact of these interventions on family and peer relationships endured beyond the end of the intervention period,[169, 179] as illustrated by a parent discussing the impact of a family art intervention:

> *I've changed because it does not have that be the first thing I tell my husband about when he calls me at night - about what's going on with (ill child) […]I've really tried to make a conscience [sic] effort not to do that….* [Parent, T1DM] [169](p. 179)

The integration of changes to family lifestyle and relationships into daily life may be an important component for maintaining the effects of the intervention on CYP's mental health over time. This will be explored further in '*Keeping it going*'. The potential improvements to family and peer relationships, in terms of increased empathy and understanding, which are

achieved through enhanced communication, may have a significant impact on CYP's mental health and wellbeing in the long term.

Summary: Social Support

The affirmational, informational and emotional support described above are all components of social support as described by Stewart and colleagues.[183] The social support received by peers may facilitate their experience of an intervention setting as a safe space, which in turn may increase the likelihood of receiving emotional and informational support. Support to share their experiences can result in feelings of reduced isolation and improved relationships with healthy peers and family. This may have important implications for extending the impact of an intervention on CYP mental health and wellbeing beyond the intervention setting. Social support seems to be an important mechanism which may facilitate the development of new skills and knowledge and help CYP create a more hopeful view for the future, as discussed below.

Construct: A Hopeful Alternative

This construct explores how the creation of a sense of hope is important to CYP with LTC and their families when considering how to reduce symptoms of mental ill-health. The first theme discusses how a sense of 'Hope and Inspiration' can be created through exposure to people who are managing to live successfully with similar conditions to the CYP. 'More than just my Illness' explains how interventions aimed at improving CYP mental health can also address other factors in addition to management of their LTC symptoms and that this is valued by CYP. 'A New Normal' considers the importance of continuing taking part in 'normal' day to day activities, whilst encouraging an acceptance of the limitations caused by the LTC. The final theme 'Mutuality' details how it is important that mental health and wellbeing interventions help CYP recognise that they too have something to offer other people in their relationships with peers and intervention deliverers.

indicates how this construct interacts with the findings of the previous two sections.

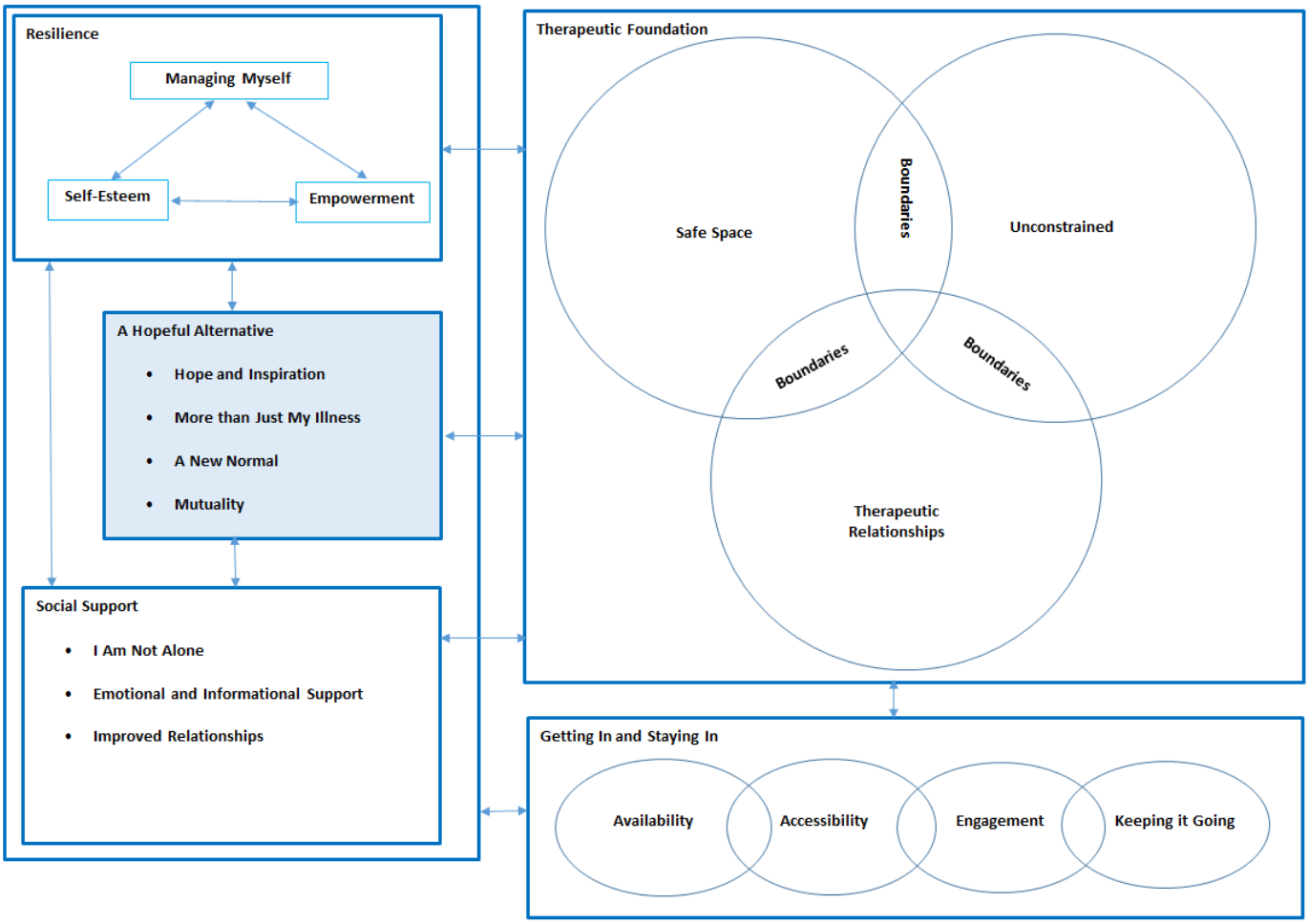

Figure 21: A Hopeful Alternative and its relationship to other constructs

Theme: Hope and Inspiration

This theme discusses how CYP value having a sense of hope whilst living with an LTC. Hope and inspiration can be provided through access to role-models who are living successfully with an LTC. Alternatively, mental health and wellbeing interventions may provide opportunities to experience success and mastery by encouraging CYP to persevere with challenging situations. A sense of inspiration may also be elicited by interventions which encourage CYP to focus on positive activities. Thirty articles contributed towards this theme (see *Supplementary Materials 2, Table 6*).

Authors of 26 studies discussed how CYP gained a sense of hope or inspiration from the intervention they were taking part in.[167, 169, 190, 206, 214, 222 168, 170, 172-174, 176-180, 186-188, 191, 192, 203, 210, 213, 224, 225] Twenty of these studies focused on CYP's interactions with peers who have an

LTC as a source of encouragement, motivation and inspiration.[167-170, 172, 173, 176-180, 186, 188, 190-192, 207, 214, 222, 225] CYP appeared to find it particularly powerful seeing an example of someone older who was leading a fulfilling life despite their illness, [167, 183, 222] as illustrated by the recipient of a peer-mentoring intervention below:

> *It was fun like it's never been before, I saw someone who is in control of the situation and isn't afraid of diabetes. If he can do it, I've got to succeed.* [CYP, T1DM] [167](p. 474)

The advice received through contact with peers of the same age within both formal and informal settings also showed that it was possible for others in the same position to overcome difficulties associated with the LTC or challenges within the intervention itself:

> *……makes you want to do something and get up and go for it. Because you see all these kids, the kids in wheelchairs and they're doing all this stuff, and they're not letting anything get in their way.* [CYP, Cancer] [178](p. 893)

> *I saw that the other girls were fine with exercises I was like ok you are worrying over nothing.* [CYP, Fibromyalgia] [191](p. 76)

Role models did not have to be someone CYP had met personally. Barlow and colleagues [176], suggest that CYP could watch vignettes of peers online to observe people who are managing to live successfully with the disease. Role models may also help encourage CYP to think about goals for other areas of their life, outside of managing their LTC, as suggested by a young woman discussing important components for a group intervention for young women with HIV:

> *Ask them who their role models are. They could be somebody famous or something like that. Be like, you see how they work hard to get that, you want to do that for yourself. You want that life for yourself.* [CYP, HIV] [173](p. 294)

This quote suggests that feelings of hope and inspiration may encourage CYP that they too can effect positive changes within their own lives outside of the interventions setting. This interpretation is supported by three studies whose authors suggest the inspiration provided by peers can also provide CYP with the motivation to persevere with intervention activities or

physical health procedures they perceive to be difficult.[189-191] This is illustrated by a parent discussing their views on the impact attending camp will have on their child:

> *I think that [friendships among children with CHD] will inspire him […] when he sees other kids do things that he might be afraid of, I think it will help him to take that leap of faith and inspire him. And he might think 'oh well let me try that'* [Parent, CHD] [190](p. 53)

The inspiration provided by peers may encourage CYP to achieve success within challenging situations. The potential feelings of empowerment and improved self-esteem which may result could be considered as important protective factors against mental ill-health, as discussed further within the overarching construct '*Resilience*' below. However, there was some evidence that CYP found talking about their emotions with peers uncomfortable[191] and some advice from peers was perceived as irrelevant if they felt that they were in a worse position than them.[176]

Authors of four studies suggest that activity based interventions which provide CYP with an opportunity to receive positive feedback, such as receiving beads following a painful procedure,[206] or provide an alternative focus through taking part in a creative activity can provide CYP with a sense of comfort and positivity.[203, 207, 210] This positive feedback may inspire CYP to continue with challenging procedural treatments and/or have a beneficial effect on their mental wellbeing.

The evidence above illustrates how hope and inspiration can be created through contact with peers with a similar illness within face-to-face, online or video settings. Interventions which provide CYP with a source of hope may also inspire increased feelings of self-confidence that they too can live a fulfilling life with an LTC.

Theme: I'm More Than Just My Illness

This theme draws on data from 33 articles (see *Supplementary Materials 2, Table 6*) and discusses how CYP value interventions which consider more than just their LTC and explores to what extent certain interventions included in the review tried to meet this need.

Eleven studies indicated that CYP valued interventions which focused on other aspects of their lives in addition to their LTC. [172, 173, 179, 181, 183, 189, 192, 200, 202, 211, 212] In addition to addressing psychosocial or educational needs[167, 176, 182, 188, 194] and acknowledging how CYP's

physical and mental health could affect one another,[191, 200, 202] interventions also created opportunities for CYP to exercise control and self-expression.[194, 203] However, there was also some evidence that some interventions failed to adequately address CYP's psychosocial needs.[176, 183, 187, 188, 222] Below a CYP describes what they would like to see as part of a group intervention for young women with HIV:

> *My perfect program wouldn't just not only [sic] be focused on the infection. It would just really be building self-worth, building self-esteem like all the way around.* [CYP, HIV] [173](p. 293)

Second-order construct data indicate CYP appeared to particularly value interventions that led to them feeling recognised as a 'normal' person,[212] and let them explore other aspects of their identities.[172, 174, 179, 211, 223] One intervention deliverer discussed how CYP used a peer support group to explore how they wished to incorporate HIV into their identity:

> *So really a means for them to figure things out on their own and figure out how they wanted to represent themselves and HIV or if they wanted that to be a part of who they were.* [Group Leader, HIV] [177](p. 387).

This suggests CYP value the opportunity to explore who they are in addition to their LTC, and services which reflect their additional needs.

Authors of five studies indicated that interventions which affect or involve other family members may also positively impact on the mental health of the CYP by addressing family interaction styles:[176, 182, 190, 194, 212]

> *My mom and dad get stressed out too and I learned some things to try to calm down when I am frustrated.* [CYP, ABI] [194](p. 659)

By considering the impact of family on CYP's mental health, it ensures CYP are not burdened with the expectation to manage their LTC and psycho-social needs by themselves. It may also help family members access appropriate support for their own needs and enable them to support their child. This could be a relief for CYP and may lead to increased empathy and understanding between family members, creating the opportunity for improved family relationships as discussed within 'Social Support'.

Theme: A New Normal

This theme explores the importance of an intervention enabling CYP to continue taking part in day to day activities, whilst accepting any limitations resulting from their LTC. The impact of CYP being able to observe how other CYP cope and adjust to life with an LTC is also discussed. Thirty-nine articles contributed towards this theme (see *Supplementary Materials 2, Table 6*).

Thirteen studies indicated CYP valued interventions which enabled them to take part in everyday activities; whether that was household tasks or doing things which friends or family without an LTC also do.[170, 172, 174, 176, 178, 182, 186, 187, 190, 191, 201, 203, 207] Below, a parent talks about the importance of a karate class her child with CP could attend with his healthy brothers:

> *There are things that go on that normal kids with no disabilities do that he can enjoy also. This helps him out the most. His brother and other kids can take karate, and so can he*. [Parent, CP] [186](p. 81)

Online interventions also aided CYP to have a link with their normal life, as illustrated by one parent who talked about how an online environment allowed their hospitalised child to engage in fun activities as they would at home:

> *Because he can't always go out and play, I think to be able to do it vicariously through the computer has been really good for him.* [Parent, Mixed] [182](p. 212)

The data above suggest that interventions which reduce CYP's sense of being different may help them feel more 'normal' and reduce their sense that they are excluded by their LTC. However, O'Callaghan and colleagues [210] suggest that some CYP may feel stigmatised by being asked to take part in a mental health and wellbeing intervention, which may act as a barrier to participation.

Eleven interventions supported CYP to accept the limitations of their LTC on their lifestyle and acknowledge that although their LTC was a part of their lives, it did not necessarily need to take over (ref 181, 184, 188, 191, 195, 201, 204-206, 222) . Below a parent discusses how camp could help their CYP to realise that having an LTC did not stop them from doing the things that they wanted to:

[interacting with children with CHD] is good for Adam to see that just because you have [CHD] doesn't mean that you can't live a full life.[…] the activities that they will do together will just help them to see that you can just be as normal as others kids…. [Parent, CHD] [190](p. 53)

In contrast to the above, parents from two studies evaluating interventions to relieve procedural distress in CYP with cancer reported that the interventions could provide CYP with the opportunity to come to terms with their own mortality. [207, 208] We suggest that for CYP with terminal illness, an intervention should focus less on creating hope, but more on achieving a sense of peace and acceptance.

Having the opportunity to meet or observe peers with similar difficulties to their own, meant CYP were able to compare the impact the LTC had on the lives of their peers and the coping strategies used by others to their own. Sometimes this encouraged CYP to develop a new, more positive perspective on their personal circumstances.[167, 169, 172, 174, 176, 178-180, 182, 183, 190, 192, 206, 207, 222, 223] This is illustrated by a CYP with JIA comparing themselves to other peers they saw in a psycho-educational video:

> *Well, I haven't got it in many places so I thought that I was lucky because other people have got it worse.* [CYP, LTC: JIA] [176](p. 602)

Despite the apparent desire to feel normal, second-order construct data from six studies suggest that at times, CYP may enjoy the opportunity to feel "special" as a result of an intervention. This may be through the recognition that an intervention is specifically for them[186, 196] or through recognition by others that they are 'brave' or 'special'.[171, 178, 206, 212] We interpret this as a possible extension on the typical developmental need to feel valued by others

Theme: Mutuality: I have something to offer to others

This theme focuses on the realisation some CYP experienced whilst receiving an intervention that they too had something to bring to their relationships with peers or those delivering the intervention. This included input on setting up new interventions or providing information, advice or empathy. This helped CYP view themselves differently, as an equal part of the relationship, instead of passive recipients of care. This has implications for CYP's feelings of empowerment and self-esteem. Thirty-three articles contributed to this theme (see *Supplementary Materials 2, Table 6*).

As later acknowledged under '*Engagement*', CYP and their families have a wealth of experience that can be called upon when designing an intervention. [173, 175, 176, 180, 189, 196-198, 202, 217, 224, 225] CYP draw upon their knowledge and experience to provide social support to their peers, [183, 222, 223] as illustrated by an adolescent discussing how they were asked to provide a peer with some informational support at camp:

> *His parents wanted me to . . . give him some advice and help him relate to how it is going to be. That really makes me feel important . . . I get to share my life experience and relate to him with his life experience.* [CYP, CHD] [171](p. 557)

CYP were able to recognise that they were able to provide support to both peers with an LTC and the intervention deliverers who had been asked to support them,[167-169, 171, 191, 223] as highlighted by a CYP talking about their relationship with their peer mentor:

> *I know him no less than he knows me, I help him, he doesn't only help me.* [CYP, T1DM] [167](p. 471)

This represents an alternate, more positive way of viewing their LTC and the impact it has on their identity and indicates that CYP can have an impact on the lives of others. We see this as a possible alternative identity that may offer a source of hope for the future for CYP and act as a protective factor against mental ill health. When CYP recognise that they too can offer beneficial advice to others it can have a positive impact on how they see themselves within the context of their relationships. No longer does their LTC mean that they are the ones who need help from others, it is also a rich source of experience that they can draw on to be 'the helpers' instead. This may improve their sense of self-esteem and empowerment and have a beneficial impact on their mental wellbeing.

Data shown in *Supplementary Materials 2, Table 7* indicate that by recognising that they too had something to bring to their personal relationships, CYP were empowered in their interactions with people outside of the intervention setting, exemplified by teaching others new skills[118] or challenging the stigma narratives around their illness:

> *It has influenced me to become more active in AIDS and HIV [sic] in teaching others about AIDS and HIV, becoming more open about my disease because I do live in a community where I have certain people around me who will not judge me for my disease.* [CYP, HIV] [177](p. 386)

The realisation that they too have something to offer others may facilitate behaviour change and CYP having a voice within the wider community. The impact of having their voice heard within an intervention is discussed under the 'Empowerment' theme below.

Summary: A Hopeful Alternative

This construct encapsulates how valuable CYP find the hope that it is possible for them to live fulfilling lives with an LTC. In addition to the importance of social support and the positive influence on peer-mentors in providing a source of inspiration, it is also important for interventions to focus on more than just CYP's physical illness. Interventions should consider how CYP can be encouraged to take part in activities which make them feel "normal", despite their physical limitations. CYP also appear to value the opportunity to recognise that they too have something to offer other people and are not just the passive recipients of help. These factors can aid CYP in feeling empowered and developing a sense of positive self-esteem; factors which may contribute towards improved mental wellbeing and discussed next under '*Resilience*' and may help CYP develop and maintain links with peers and family, as discussed previously under '*Social Support*'.

Construct: Resilience

This construct conceptualises resilience as a process which occurs through CYP's involvement with an intervention and interaction with the environment/systems around them, which influences how CYP perceive themselves and their abilities. The themes described below focus on issues which may influence CYP wellbeing, in addition to potentially impacting on clinical symptoms of mental ill health. The first of the inter-related categories 'Managing Myself' discusses how CYP benefit from interventions which equip them with skills and knowledge to help them manage their emotions and relationships and take responsibility for managing their LTC. This can help them feel empowered and develop positive self-esteem. The second theme discusses how being given opportunities for choice and encouragement to be more involved in their care is key to develop feelings of 'Empowerment'. The third and final theme discusses how a positive sense of 'Self-esteem' may be promoted through positive interactions with others and experiences of mastery; which in turn further promotes the use of positive coping strategies. *Figure 22* shows the relationship between 'Resilience' and the other constructs in the synthesis.

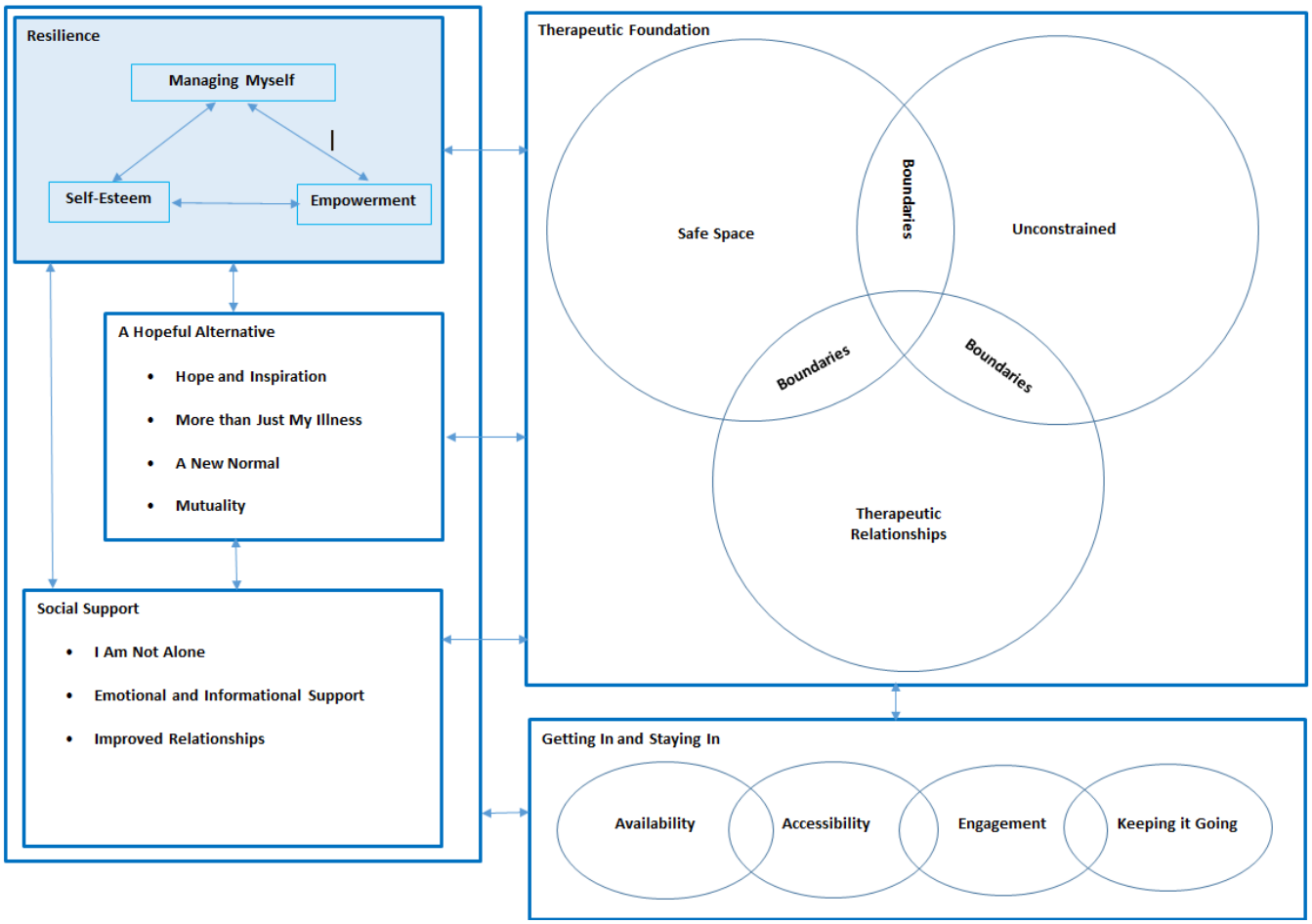

Figure 22: Resilience and its relationship to other constructs

Theme: Managing Myself

This theme explores how being taught skills and knowledge to manage both their physical and mental wellbeing can enhance CYP's ability to cope with difficult life circumstances, and lead to a sense of empowerment and improved self-esteem. Forty-nine articles contributed to this theme (see *Supplementary Materials 2, Table 6*).

Twenty-three studies indicate that CYP seemed to find it helpful to have the opportunity to learn skills which would allow them to take responsibility for managing their LTC as part of an intervention aiming to improve their mental health. [118, 167, 176, 183] [168, 171, 173, 177, 179, 181, 182, 187-189, 191, 195, 196, 198, 200-203, 220] Such learning appeared to take place through interventions in one-to-one, group and online settings and increased CYP's confidence to manage their LTC in the future: (ref 197, 205, 217)

Learning the techniques helped my asthma and me as a person, because if I know how to control my asthma now, I can help in the future and stop things from occurring [CYP, Asthma] [118](p. 893)

One CYP receiving CBT described how the skills they learned provided them with a sense of control over their fibromyalgia:

It gave me something to work with which is something I haven't had since I was in school. [CYP, Fibromyalgia] [187](p. 176)

Three studies highlighted how learning how to manage LTC and what to expect in the future could help them feel prepared and increase feelings of self-efficacy, mastery and being in control.[173, 176, 182] This may have important implications for helping reduce CYP's anxiety around not knowing if they will be able to cope with physical, emotional and social challenges arising from their LTC.

CYP and their parents also appreciated skills learnt during mental health and wellbeing interventions that directly enabled CYP to manage stress[118, 167, 176, 182, 188, 189, 194-196, 201, 202, 206, 211] and in some cases showed that they were able to use these new skills when required during their day to day life, [183, 187, 191, 195, 201, 222] as illustrated by two participants of a neuromuscular and CBT training programme:

Mini-relaxation I would do all the time when I felt a little tense... muscle relaxation I would usually do in the morning or at night before I went to bed. [CYP 1, CFS]

Pleasant imagery I usually did before bed or if I was having a lot of pain...calming statements...I did mostly when I felt...anxious or when I couldn't sleep. [CYP 2, CFS] [191](p. 74)

Interventions which acknowledged the interaction between physical symptoms and mental wellbeing seemed to be appreciated,[202] particularly for LTCs such as fibromyalgia and CFS. [191, 200]

Acquiring skills to manage challenging social situations seemed to be particularly important for CYP living with LTCs associated with a high degree of stigma, such as HIV, where there were issues around managing intimate relationships and disclosure.[173, 181, 192]

Hosek and colleagues[173] highlighted a demand for group interventions which support young women who have HIV to manage the emotional distress resulting from negative consequences following disclosure:

> *It doesn't mean that because I told my friend that I'm HIV positive she's gonna accept me. It doesn't mean that because I told her I have to accept that she has to accept me. […] She's out of my picture. So how do we deal with it.* [CYP, HIV] [173](p. 294)

Eleven studies included in this review indicated how distraction can be a valued skill CYP can use to cope with distress and pain and escape the boredom or uncertainty associated with long hospital stays.[169, 174, 178, 179, 182, 196, 207, 208, 211-213] The evidence presented within this theme suggests that gaining skills to managing their LTC, social situations and distressing procedures through a mental health intervention, can help CYP to take responsibility for managing their physical and mental wellbeing.

Theme: Empowerment

Forty-seven articles contributed to this theme (see Supplementary Materials *2, Table 6)*., which discusses how mental health and wellbeing interventions can empower CYP to take responsibility for their own health care needs within day to day and healthcare settings, have a voice within their local community and increase confidence in managing tasks of everyday living. CYP can feel empowered as a result of an intervention which includes the incorporation of activities which allow CYP to experience mastery and achievement, and having their voices heard within an environment which encourages reflection on progress. The experience of feeling empowered within the intervention setting can impact on how able CYP feel to affect change within their day to day lives and is thus an important component of resilience. Feeling empowered can also affect how CYP view themselves in relation to others and thus relates to the '*Mutuality'* theme above. Other third-order constructs which contributed towards this theme can be seen in *Supplementary Materials 2, Table 8*.

Evidence from 11 studies suggest interventions can offer opportunities for CYP to make choices and exercise autonomy by deciding how they would like others to interact with and

support them. [167, 175, 178, 182, 183, 188, 190, 200, 203, 204, 213] An example of this is provided by a parent describing how their child instructed staff on how best to support them whilst they were undergoing needle insertions:

> *(The child) wrote out a needle plan, and she talked it through at home and worked it out with the psychotherapist here at the hospital. […] every time there was a new nurse or doctor she'd hand the card to them and they had to read it…* [Parent, CF] [204](p.336)

It appeared to be important for interventions to provide opportunities for CYP to experience having their voices heard. This could be achieved through allowing CYP to vote on important decisions as part of the wider group and the chance to tell their story,[176, 178, 181, 186, 203, 204, 206, 208] These experiences enabled children to make their voices heard within the wider community.[169, 177, 181] One parent observed how a camp for CYP with CHD allowed their child to develop confidence in their ability to speak up for what they neededlater whilst attending school:

> *Since going back to school (…)she knows that from camp as well, that she has to be an advocate for herself. That she's the one that has to kind of put the foot down and say 'this is why I need to stop and I'm going to stop'* [Parent, CHD] [190](p. 65)

Data from Supplementary Materials 2, Table 7 also indicates that activities incorporated into interventions which promote recognition of achievement and mastery of challenges may help CYP gain a sense of confidence in their abilities.[118, 170-172, 174, 176, 178-180, 182, 184, 190, 191, 204, 206, 207, 211, 212] This is supported by a quote from a an intervention deliverer from a camp intervention:

> *For some of these people, they're very sheltered and their parents don't let them do anything, and they're climbing rock walls and going down zip lines, and the fact is that they're overcoming their fears and obstacles.* [Intervention deliverer, Cancer] [178](p. 893)

Having this sense of confidence in their own abilities may enable CYP to feel empowered outside of the intervention setting. Some of the outcomes which were more indirectly related to mental health included the willingness of CYP to become more involved in their care, [204] to take responsibility for completing homework exercises[118] and complete

other tasks in order to manage their health needs.[118, 167, 176, 181, 183, 189, 190, 192, 200, 203, 222] For example, in a study which examined the effectiveness of breathing re-training for asthma, one adolescent reported that it was they who took responsibility for remembering to do the exercises:

> *Nothing has gotten in the way, I just remember, no one reminds me.* [CYP, Asthma] [118](p. 893)

Overall this theme illustrates how mental health interventions which offer opportunities for CYP to exert control, gain experiences of mastery and feel that their voices are heard, can enable them to feel empowered within their everyday lives beyond the intervention setting. This may contribute towards CYP's development of 'Resilience' and a positive view of themselves.

Theme: Self-esteem

This theme details how improved self-esteem may contribute towards the development of resilience. A CYP's self-esteem may be improved by an intervention that encourages the child to recognise their capabilities and areas of achievement, as well as providing opportunities to interact with others who can reflect the positive parts of their identity. Twenty-seven studies contributed to this theme (*Supplementary Materials 2, Table 6*).

Data from four studies indicate that taking part in those interventions reinforced CYP's belief in their own ability to manage their condition, emotions and negative responses from other people.[183, 190, 203, 212]

Authors of six studies suggested that CYP valued the opportunity to recognise positive things about themselves. [169, 173, 184, 191, 194, 195] One study encouraged CYP to practise making positive affirmations about themselves during the day, and there was some evidence to suggest that CYP could then use this to ameliorate emotional distress in other settings:

> *Thought about what I was proud of or what made me happy, which helped me to calm down and be happy again.* [CYP, T1DM] [195](p. 482)

The above quote demonstrates the impact of CYP having the opportunity to learn skills to manage their emotions on their sense of wellbeing and suggests opportunities to build CYPs self-esteem can provide a valuable resource for CYP to draw upon in times of emotional need.

Authors of 17 studies suggested that positive recognition and affirmation from peers, family, intervention deliverers and themselves was found to be important for improving CYP's self-esteem and/or mood.[167, 169, 171, 178, 183, 184, 186-188, 190-192, 194, 202, 206, 207, 212] One parent felt their child found it valuable to recognise that he was capable of offering support to peers through a computer-based intervention and that other people also recognised this:

> *I think it encouraged [the ill adolescent] ...to know that he had the ability to 'do.' I think it really boosted his confidence, knowing that he had something to offer other kids, and that someone saw that while he was on the computer.* [Parent, Mixed] [182](p. 213)

Authors of three studies indicate opportunities for CYP to tell their story to others, helped CYP communicate their experiences and receive positive recognition from others.[206, 207, 212] Kashikar-Zuck and colleagues [191] indicate CYP appreciated the tailoring of intervention tasks to their individual abilities, alongside the opportunity for progression through stages of increasing difficulty. One quote suggests this structure provided CYP with the chance to reflect back upon their achievements with pride:

> *I am glad that I went through it... now I can look back...at how far I have come... today that stuff, wasn't hard. It just gives you this sense of pride, like if I can overcome that, what else can overcome?* [CYP, Fibromyalgia] [191](p. 77)

The importance of the individual delivering an intervention in being able to relinquish control within an intervention setting to help CYP recognise that they too have something to offer is highlighted within *Supplementary Materials 2, Table 7* under the 'Empowerment' theme. The below quote highlights the emotional impact of when CYP perceived an intervention deliverer as being unable to acknowledge their views as equal to their own:

> *I spoke to (therapist's name) about it, […]and I said to him that it wasn't working and I didn't know why, he basically told me it was my fault and that if it wasn't working it must be because I wasn't doing it properly*. [CYP, CFS] [200](p. 519)

This quote suggests the CYP was left feeling blamed and criticised when they felt the intervention deliverer had not respected their views. This highlights the need for empathetic, supportive therapeutic relationships with intervention deliverers as discussed within '*A Therapeutic Foundation*' and the potential impact of these relationships on CYP's self-esteem.

Second-order construct data from four studies indicates that having the opportunity to reflect on what they have been through may be meaningful for CYP. Such reflection appeared to be facilitated through the receipt or creation of "mementos" documenting their LTC and treatment story and chance to observe change over the course of an intervention[172, 178, 179, 206] Combined with the positive responses these mementos elicit from others, they may reinforce CYP's self-concept as someone who is brave, special and resilient. This may bolster their sense of self-esteem which could be a protective factor against the development of mental ill-health.

Having the opportunity to achieve a sense of mastery by successfully completing activities not only empowered CYPs as described above, but also improved their mood[186, 191] and/or increased their sense of self-esteem:[172, 182, 184]

> *I have to say my favorite [sic] memory from camp would probably be climbing the tower. It was an incredible experience. When I got to the top I felt like I accomplished, you know, what I wanted to do. The zip line was probably the best part also.* [CYP, Mixed] [172](p. 116)

Quotes from five studies suggest that interventions which encouraged focus on areas of ability within the individual child, rather than what they were not able to do, were also valued.[171, 173, 186, 190, 207] This is illustrated by a parent describing the importance of a karate class to provide an example of what they could do for their CYP with CP:

> *I don't want his sister or brother to be in a private karate class. This has really helped him a lot. Even during the week when they do things that he can't do, we mention the karate thing. I tell him that he does karate and they don't.* [Parent, CP] [186](p. 82)

Opportunities which allow CYP to feel capable and included may boost CYP self-esteem by supporting them to develop a more hopeful view of their capabilities and of the future, as described under '*A Hopeful Alternative*'.

This section discusses the role of mental health and wellbeing interventions which encourage positive recognition and experiences of mastery in boosting the self-esteem of CYP. This may result in CYP having a more positive image of themselves, which may act as a preventative factor against experiencing symptoms of mental ill-health.

Summary: Resilience

This theme explored three inter-related components which may promote resilience within CYP with LTCs. 'Managing Myself' detailed how it is necessary that CYP are supported to gain the relevant skills and knowledge they need in order to be able to take age-appropriate responsibility and have a voice within the intervention process and the wider community. This knowledge transfer can occur through both formally "taught" interventions and by more informal social learning through contact with peers in group interventions. Having the appropriate skills may facilitate the development of 'Empowerment'. Here the importance of allowing CYP opportunities to make choices about the care they receive and take responsibility for managing the different areas of their lives affected by their illness was explored. The recognition of both their success in managing their LTC and achievement in other areas of their life facilitates a sense of positive self-esteem, the third theme conceptualised here as being necessary to promote resilience.

Construct: Getting In and Staying In

This construct details the different continuous/overlapping stages as a CYP accesses an intervention; from the development of interventions aiming to improve the mental health and wellbeing of CYP with LTCs, through to CYP being able to access and stay engaged with the intervention. 'Availability' explores how issues such as the initial recognition of mental health need and provision of required resources are necessary for an intervention to be considered. The different factors required to ensure CYP and their families can access these interventions is discussed under 'Accessibility'. Issues which may affect ongoing engagement with mental health and wellbeing interventions are detailed within 'Engagement'. The final theme 'Keeping it going' discusses the need for interventions to have components which can be adopted into everyday life, ensuring that beneficial effects are not confined to within intervention sessions. The inter-relationship between 'Getting In and Staying In' and the other four constructs is shown below in *Figure 23*.

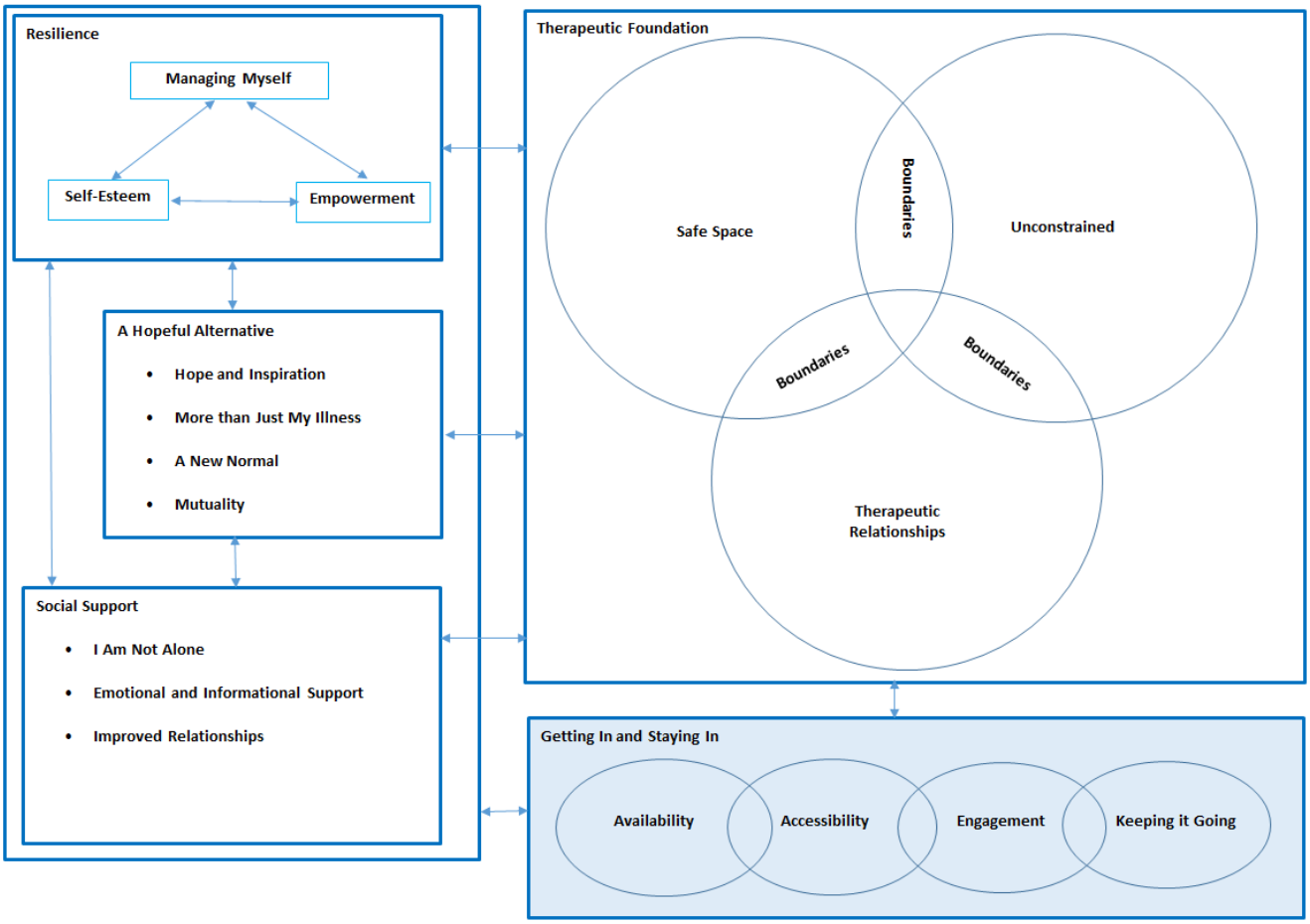

Figure 23: Getting In and Staying In and its relationship to other constructs

Theme: Availability

This theme draws on 30 articles (see *Supplementary Materials 2, Table 6*) and centres on the need to recognise the mental health needs of children with an LTC and ensure the availability of relevant resources to address them.

Authors of three articles acknowledge the difficulty for health services to identify CYP and/or their families in need of support to maintain their wellbeing. [167, 176, 192] One parent expressed frustration at the difficulty of getting the mental health needs of their child recognised by health professionals focusing on physical symptom management:

> *They really don't understand the problems you have at home...The doctors see them for half an-hour...and examine the joints...but, it doesn't just affect their joints. It affects them mentally and that's what we have to put up with.* [Parent, Juvenile Idiopathic Arthritis (JIA)] [176](p. 602)

Participants and authors of seven studies commented that when mental health interventions were provided, the availability or range of options offered was sometimes limited:[173, 176, 180, 181, 191, 194, 203]

> *[The ChIPS intervention] wasn't easy to find. I was looking for some support for [my son] because he was struggling last year […]… They said they only had one social worker for the outpatient department and it was all very difficult for them to see him.* [Parent, Mixed] [180](p. 2546)

Authors of two studies highlight the need for staff expertise in order to design and set up an intervention[176, 202] and ensure a suitable space for the intervention to take place within is available.[181, 202] Authors of a further nine studies highlighted the need to ensure that staff delivering the intervention had the appropriate skills, experience and training so that the intervention was delivered as planned and that the emotional or behavioural needs of the CYP were met. [167, 171, 176-179, 183, 194, 203]

Second-order construct data from three studies suggest that when mental health and wellbeing interventions are available, some CYP may not feel able to seek support for their wellbeing. This may be for a variety of reasons; whether this was through a desire not to discuss their anxieties or their LTC in front of parents, [167] fear of physically hurting someone by holding their hand whilst undergoing painful procedures,[213] or the view that the setting in which they were being seen was not appropriate to their perceived needs. [187]. Overall, this theme highlights the need for CYP's mental health needs to be recognised by staff they may come into contact with and relevant interventions to be provided to CYP in a timely manner..

Theme: Accessibility

This theme focuses on factors that may affect whether CYP and their families are able to access an available intervention. Factors that may facilitate access include; pre-existing relationships with health and social-care staff, ensuring interventions consider the individual needs of the CYP and addressing practical issues such as intervention setting, duration and timing are convenient. Fifty-two articles contributed to this theme.

Seven studies suggest that both health and social care professionals play an important role in identifying CYP who may benefit from existing interventions, ensuring families are

aware of the support available and inviting them to participate in the intervention.[171, 174, 177, 180, 182, 192, 203]

Approaching CYP directly via text message or letter was an effective means of supporting them to access an intervention,[180, 192] whilst CYP may observe others taking part and wish to participate.[177, 182, 187, 211] There was suggestion that more formal processes could ensure interventions were accessible by those who needed them, including introduction of referral systems[203, 217] and ensuring that CYP from certain ethnic minorities or with increased illness severity are able to attend. [174] Parents felt that the means of introducing mental health interventions relevant to their child should be incorporated into inpatient discharge processes:

> *You've got the teachers, you've got the wards, you've got the social workers. It should be just part and parcel of coming around on discharge, you know that you are aware that there is support for young people, maybe not now, but when it may be appropriate.* [Parent, Mixed] [180](p. 2547)

The idea that the unpredictable nature of an LTC can prevent CYP from accessing, or participating fully, in treatment for their mental health was recognised by 14 studies.[169, 170, 178-180, 182, 187, 188, 190, 191, 196, 207, 208, 211] Therefore, the recognition of physical health needs by those planning mental health and wellbeing interventions and consideration of how the intervention can be altered accordingly is required to ensure that the intervention is accessible by those who need it.[178]

By ensuring CYP's physical health needs are met, intervention deliverers and services can reduce some of the anxiety around attending an intervention, which may help it to be perceived as a 'Safe Space' for CYP and their families.

Facilitators in online interventions may also play an important role in stimulating discussion and ensuring the relevance of the material to those taking part.[183, 202, 222] This may help CYP who are less confident access intervention material and maintain their engagement. Six studies suggested that the communication skills and understanding of those delivering the intervention was also key in making the material accessible.[167, 171, 190, 191, 200, 206] One CYP highlights how they felt the person delivering the intervention did not understand the material and this meant that the information delivered was not fully understood:

The physiology stuff at the beginning I didn't find helpful because I don't think it was fully explained, cause to be honest I don't think they fully understand it themselves. [CYP, CFS] [200](p. 518)

This has important implications for CYP acquiring the skills and knowledge required to manage their physical and mental wellbeing as described under '*Managing Myself*'. First and second-order data in *Supplementary Materials* 2, Table 7 suggest that interventions within this synthesis used different ways to ensure that their material was accessible through considering the age, educational and developmental level of the CYP.[118, 169, 173, 176, 179, 187, 189, 192, 196, 202, 203, 206, 207]

Second-order construct data from 13 studies also suggest that the timing of the intervention and choice of venue need to be considered to ensure that it does not interfere with CYP's education or parents' employment and that the duration and financial burden of travel does not prevent attendance.[118, 169, 177, 180, 181, 187, 188, 191, 192, 196, 200, 207, 222] The usefulness of coordinating delivery of physical and mental health interventions was highlighted [181, 196], and is illustrated by a quote from a young person who had attended a hospital-based support group:

It's convenient and just you can do everything all in one day and see everybody in one day, and just get everything done. [CYP, HIV] [181](p. 8)

We suggest that considering CYP's physical, psycho-social and family needs together may not only reduce the burden of attending multiple appointments, but also reinforce that CYP and their families have other needs in addition to their LTC and are part of a wider-family system as discussed in '*More than just my illness*' . This may help CYP feel validated as a whole person and make them feel supported and understood by services, necessary components to build 'A Therapeutic Foundation'.

Interventions delivered online can overcome some of the practical issues around access. One of the perceived benefits of such interventions was that they enabled children to access the intervention independently and overcome the physical limitations of their illness, although sometimes technological issues hindered this:[182, 183, 188, 222]

The biggest (challenge) is consistency with working the computer games. Often (a window with a picture of) a dog comes up and says that they're unable to locate their site or for safety purposes it's been blocked. And a lot of the time they go to

put on a game and they can't get it to work. So that gets frustrating. [Health Care Provider, Mixed] [182](p. 218)

However, authors of six studies suggest CYP and their families would like flexibility in terms of how an intervention is delivered.[173, 183, 184, 194, 195, 202] Parents acknowledged the convenience of interventions supported by technology, but stated that they would also like CYP to have the opportunity to meet peers face-to-face and/or intervention deliverers face to face.[183, 194, 195, 222]

Care is required that the intervention itself is not too burdensome for CYP or their families. Authors of four studies suggest that the pace and duration of intervention settings should be considered to ensure that they are not too long, yet allow enough time for the material to be covered without overwhelming participants.[169, 183, 191, 200] An example of a situation where the long duration of the session meant that the CYP found it too burdensome is shown below:

It was[…] really taxing the first day because it started at 10 and didn't finish until about seven in the evening, and there were also about seven other people there which is like…considering I had been like housebound for years I hadn't really got…like, saw anyone,[…] I was absolutely shattered. [CYP, CFS] [200](p. 517)

This quote suggests that a balance is needed between the convenience of holding multiple appointments at the same time and ensuring that CYP and their families do not feel overwhelmed.

The perceived burden of sessions may be impacted by the type of LTC experienced by CYP as well as the stage of their illness. Studies in this review support this idea by discussing the perceived importance of ensuring that a mental health and wellbeing intervention acknowledges that the LTC may impact on the mental health of CYP and address this by considering LTC specific challenges or worries [171, 172, 176, 178-180, 187, 189, 191, 192, 194, 203, 211] and stage of illness.[176, 183, 185, 189, 192, 194, 200, 203, 207, 211, 223] An example of the role of the intervention deliverer in ensuring exercises and activities are within the physical capabilities of CYP is given below, within a cognitive-behavioural neuromuscular training programme for CYP with fibromyalgia:

(The exercise physiologist) would work with you personally to make sure you were comfortable with where you were…make sure the exercise wasn't too easy or too hard [CYP, Fibromyalgia] [191](p. 74)

The role of the intervention deliverer in establishing boundaries to ensure that participants feel safe enough to discuss emotive subjects is discussed within '*A Therapeutic Foundation*'. Having opportunities to chat before the interventions and including activities which CYP and their families found enjoyable also appeared to make the more formal therapeutic aspects of intervention accessible.[169, 210] One example is the art activities during a family art intervention seemed to provide an alternative focus and aid disclosure:

Researcher: Was Living Well what you expected it to be?

Parent: Actually, it wasn't. And that's good. I thought about everybody talking about their kids would be kind of depressing, but it turned out, the artwork and everything kind of opened everybody up. [Parent, Mixed] [169](p. 188)

Authors of five studies acknowledged that the interventions being evaluated were not able to address CYP's every need or replace other interventions which may be useful.[183, 187, 188, 194, 202], this highlights how CYP with LTCs have other needs in addition to their physical health, as discussed under '*More Than Just My Illness*'.

This theme summarises different factors that may influence how accessible CYP and their families find an intervention and the role of the intervention deliverer in adapting the intervention to ensure that it meets CYPs psycho-social needs.

Theme: Engagement

Whilst the availability of an accessible intervention is necessary to allow for it to be provided, CYP may not necessarily want to engage. Different factors which may influence engagement with an intervention are explored next.. Fifty-four articles contributed to this theme (see *Supplementary Materials 2, Table 6*).

The role of the intervention deliverer in adapting the intervention to ensure its relevance to CYP is discussed in above within '*Accessibility*'. *Supplementary Materials 2, Table 7* indicates that there are multiple ways in which mental health and wellbeing interventions can be flexibly delivered according to the needs and individual characteristics of CYP. Authors of

six studies suggest that interventions which considered the individual behavioural, social and emotional needs were valued by CYP and their families. [173, 178, 184, 186, 187, 208] Interventions included within the review also considered stage of illness [176, 192, 194, 200], gender[167, 173, 174, 189, 203, 211, 223] and the individual interests of CYP taking part in the intervention [167, 177, 182, 187, 196, 207, 212] as factors which could ensure a mental health intervention's relevance to CYP and help sustain their engagement with the intervention.

In contrast, authors of seven studies suggest CYP can engage with an intervention at a level which meets their needs, sometimes with no formal adaptation of the intervention process. [167, 168, 172, 179, 182, 188, 212] For example, Gillard and Allsop [172] discuss how a camp intervention appears to meet differing needs among CYP, as influenced by their type of LTC. This fluid interaction of CYP with the interventions may be because the design of the interventions allows for it. CYP can choose which aspects of the intervention are relevant to their needs and access the intervention at this 'level'. Not only can this be empowering for CYP, as discussed above, it may also mean that some interventions could meet the mental health needs of CYP as they change over time. This idea is explored further within '*Keeping it Going*'.

Explaining the rationale or theory behind an intervention can also facilitate the engagement of CYP with an intervention.[172, 200, 211] One CYP discusses how helpful they found understanding the rationale behind an intervention to treat their CFS:

> *The explaining of the process, the background information definitely helped because it helps you realize how the process works, and there is no point doing something unless you understand how it works… otherwise you won't believe in it enough.* [CYP, CFS] [200](p. 518)

Both CYP and their families appeared to engage with interventions which were evidence based or developed through consultation with other families, as this appeared to give the interventions validity and made them worth 'buying in to':

> *This would be a very good tool. (This has) techniques developed through research with other families and (would be) very beneficial* [Parent, Cancer] [196](p. 3321)

The above suggests that CYP and their families feel able to trust and perhaps put their faith in interventions that are tailored to meet their needs and 'make sense'. This could be

considered an important part of knowing what to expect from an intervention, as discussed earlier under '*Safe Space*'.

Second-order construct data from *Supplementary Materials 2, Table 7* indicates that CYP and their families can have a variety of different expectations prior to the beginning of an intervention[187, 200, 201] and the extent to which these expectations match their experience of the interventions may affect how effective the intervention is perceived to be.[200] CYP expressed anxieties about encountering new health professionals, not knowing anyone else or what to expect and a fear of not knowing what to say to someone who was going through difficult experiences or dying. [168, 169, 187]

Most CYP's concerns appeared to dissipate over the course of the various interventions.[180, 184, 186, 191] Some CYP and authors of studies believed that an initial tolerance of anxiety, discomfort or frustration was necessary to both begin and maintain engagement with an intervention:[176, 177, 182, 191, 203]

> *Don't be shy because if you're shy you miss out on a lot.* [CYP, HIV] [177](p. 387)

This contrasts with the idea explored in the '*Safe Space*' theme that familiarity with the person delivering the intervention or peers is required before change can occur.

First and second-order construct data from *Supplementary Materials 2, Table 7* suggest that fun or pleasant activities appeared to be important in motivating CYP, particularly those who were younger, to engage with an intervention.[167-173, 177-179, 182, 183, 186, 190, 192, 194, 202, 203, 207, 211, 212, 222] Other components that enhanced engagement with, or adherence to, an intervention included the provision of a meal during the session, the receipt of rewards or "gifts", and the opportunity to keep a product of the intervention as a memento or display it for others to see. [169, 178, 179, 192, 195, 206, 207, 217]

Some evaluations of online support interventions acknowledged the importance of clear website layout and attractive graphics to maximise user engagement.[182, 195, 224, 225] A quote from a CYP evaluating an online self-management programme for CYP with JIA, emphasises how large amounts of text can be off-putting for young people:

> *Like this, it looks like a lot of stuff to read, and I probably, as a teenager…wouldn't take the time to read it.* [CYP, JIA] [224](p. 10)

Information from six studies indicates that many families appreciated telephone calls or texts from intervention deliverers or researchers to remind them to attend appointments, carry out the intervention at home or use the intervention when required.[182, 191, 195, 196, 202, 207] Three additional studies suggested that CYP and their families may benefit from information that they can refer back to at the end of an appointment or intervention.[118, 176, 194] In addition to facilitating engagement with an intervention, these memory aids can also help CYP and their families to incorporate advice and skills practise into their daily lives. This may extend any potential benefits of a mental health and wellbeing intervention beyond the scheduled end of the intervention, a concept which is discussed below within '*Keeping it going*'.

Theme: Keeping It Going

The content of this theme draws upon 42 articles (see *Supplementary Materials 2, Table 6*) and discusses the importance of ensuring interventions are adequately resourced and have components which can be adopted into everyday life by CYP and their families. Some interventions may encourage the development skills such as emotional management by CYP, which may increase the likelihood that any benefits CYP experience to their mental health and wellbeing achieved can be sustained beyond the end-point of an intervention.

The importance of ongoing training or supervision with peers or colleagues with specialised mental health training, particularly for intervention deliverers with non-mental health backgrounds, was recognised by authors of three studies.[167, 183, 203] A higher degree of training was observed for individuals delivering more specialised mental health interventions, such as art therapy.[177, 194, 203] Without such ongoing support, it would be difficult to sustain delivery of an intervention long term and maintain engagement with CYP by ensuring the intervention meets their mental health and wellbeing needs.

The need for training prior to the onset of an intervention to ensure intervention deliverers were able to meet the psycho-social needs of CYP taking part was implied in a further three studies.[171, 177, 206] This is illustrated by a quote describing how intervention deliverers needed to be able to respond quickly to manage the disclosures of CYP during a group creative writing intervention:

> *And then there's the risk that a kid starts to write something that maybe puts them in a precarious situation that we have to address in the class. We didn't know*

> *what was going to come out, what kids were going to write, what they were going to say.* [intervention deliverer, HIV] [177](p. 387)

This interpretation is closely linked to the need to provide appropriately trained staff to deliver an intervention, as discussed under '*Availability*' above. The skills of individual intervention deliverers and the support provided to them may determine how well an intervention of longer duration can meet the needs of CYP as they change over time. Manualized protocols may also help facilitate the training of health/social care staff and consistent delivery of mental health interventions.[191, 194, 206]

Financial support to buy appropriate equipment which CYP could use if required[178, 182, 211] and fund staff time[194, 196, 204, 212] was also recognised as essential in ensuring that an intervention was sustainable. One CYP from a family art-therapy intervention indicated their concern regarding issues relating to the funding of an intervention:

> *I worry that they're not going to get funded because there might not be a huge change in someone, but when you're chronically ill, you don't need a huge change to make a difference; […]I think whoever grants you all the money needs to know that…* . [CYP, Mixed] [169](p. 238)

The above quote hints at the need for services to prove their efficacy in order to receive funding and the potential impact on the mental health of CYP who are aware that funding for an intervention they find helpful may not be secure.[194] Resource and funding issues may have implications for the ability of intervention deliverers and services to provide an intervention which can be experienced as a 'Therapeutic Foundation' by those who access it.

Data from 16 studies suggest that over time, the psycho-social impact of a mental health intervention may be seen outside of the original intervention setting, as discussed previously within '*Improved Relationships'* and '*Managing Myself*'.[167, 171, 172, 177-180, 182, 183, 190, 191, 201, 203, 211, 212, 223] One factor which appeared to facilitate this included the opportunity for CYP to practise the skills they have learnt, both within an intervention setting[191, 200] and within their everyday lives.[191 118, 169, 196, 200] One CYP describes how having materials to support practise of relaxation skills was helpful for encouraging practise outside of a formal intervention settings:[118, 194, 195]

I found the CD to be most helpful. I practiced twice a week. Will still use it even after the study [CYP, LTC: Asthma][118](p. 893)

Authors of ten studies suggested that technology, including social networking sites, can provide the opportunity for CYP to continue supportive relationships developed during peer based interventions.[168, 170, 171, 174, 179, 180, 184, 188, 190, 223] A quote from a CYP indicates how these relationships can endure across time, despite relatively little face-to-face contact:

She was the one I called when I got cancer. I don't know. Even though like we never see each other, I feel like she is one of my closest friends. Like, closer than the ones I have at school. [CYP, CHD] [170](p. 545)

Authors of six studies suggest that intervention deliverers can play a role in extending the impact of the intervention beyond its time-limited delivery period, by creating opportunities to meet again after the intervention has ended[171, 179] or by providing "top up" intervention sessions based upon the changing needs of the child.[176, 194, 200, 211]

Positive feelings regarding interventions can permeate CYP's lives. Six studies suggest that merely the memory, facilitated by mementos or anticipation of the intervention, could have a positive impact on their feelings of wellbeing for both the CYP and their family: [171, 172, 178, 179, 206, 209]

The meaning of camp in my life is everything. It's what I want to do every single summer. I look forward to it every single summer … So it's really, really fun. [CYP, LTC: Mixed] [172](p. 115)

I'll tell you that it's something we won't forget. It will be a memory that we will share. Together, the four of us. And you know, we talk about things that we did at Living Well. [Parent, LTC: Mixed] [169](p. 172)

Four studies suggest that having a memory from the intervention to think about, or an object, such as a toy, to use can be a source of comfort for CYP, particularly those undergoing distressing procedures. [179, 196, 206, 211] These physical objects or memories can act as a reminder to CYP of their resilience by reminding them of how they have successfully overcome challenges within their physical health treatment and within an intervention setting.[179, 206] These

'transitional objects' may act as a source of emotional comfort[196, 211] and be utilised in times of stress to improve emotional wellbeing.

Summary: Getting In and Staying In

"Getting in and Staying In" acknowledges that getting CYP involved with an intervention that may benefit their mental health is a process, requiring the availability of an accessible intervention, which is both tailored to the needs and characteristics of the CYP it intends to support, but is also capable of adapting over time if their needs change. To ensure that an intervention is sustainable, adequate resources must be available to both implement the intervention and continue delivery over time. Interventions demonstrate sustainability where CYP are using aspects of the intervention or developing relationships outside of sessions. It appears that both parents and service professionals have an important role in supporting CYP to access and engage with an intervention and encourage long term behaviour/emotional change.

Line of argument synthesis

The line of argument presented in aims to illustrate the relationships between the different themes discussed within this chapter. Overall it describes the process of CYP with LTCs accessing and maintaining engagement with a relevant mental health and wellbeing intervention, acquiring a sense of hope for the future and increased resilience, all against the backdrop of a safe space with adequate social support. However, this relationship is tentative and does not necessarily mean that increased experience of a safe space and social support leads to increased hope and resilience as other factors may modify this relationship.

The process illustrated within 'Getting In and Staying In' shows CYP's journey from the recognition of their mental health needs to being offered an available intervention, through potential barriers to accessing an intervention and continued engagement if appropriate. Maintaining engagement with an intervention and the experience of an intervention setting as a 'Therapeutic Foundation' seem to be closely interlinked. In particular, the aspects of the 'Engagement' and 'Safe Space' that encourage addressing worries prior to the intervention and the use of a familiar routine and structure within intervention settings in order that CYP know what to expect are inter-related. The more CYP engage with a setting, the more familiar that setting becomes; thus reinforcing both the potential for continued future engagement and the

experience of that setting as safe. Not only does this have a positive impact on CYPs anxiety about meeting new people, but it also allows them to develop relationships with empathetic intervention deliverers and peers; which can aid the CYP in telling their stories and expressing their feelings to an empathetic audience. This process of speaking and being heard can allow CYP to access the affirmational, emotional and informational support detailed under the social support theme, which can further reinforce the experience of a setting as being a safe space. This opportunity to discuss feelings and experiences can be a positive release. Evidence indicated that CYP highly valued the sense of reduced isolation which accompanied the chance to meet peers with similar experiences to themselves.

Alongside the relationships being developed within the intervention setting between CYP, their peers and those delivering the intervention, the informational support acquired as a result of these relationships also enabled CYP to acquire skills to address challenges in their relationships with healthy peers and their family. The resulting improved relationships between CYP, their families and both peers with an LTC and those without, may contribute to the continued engagement of CYP and their family with an intervention and maintenance of the effects beyond its scheduled end.

Having access to a 'Therapeutic Foundation' also allows CYP to utilise the informational support offered by peers and intervention deliverers to develop new skills and knowledge to manage their LTC and emotions. This can help facilitate feelings of empowerment, as can the experience of mastery of tasks both related and unrelated to managing their LTC. These experiences of mastery, alongside encouragement to recognise positive aspects about themselves can also contribute to feelings of improved self-esteem and empowerment. As indicated within , this inter-related set of themes can contribute overall to the resilience of the CYP and their ability to cope with both their LTC and mental health difficulties. In addition to increased resilience, having the opportunity to observe other people who are able to successfully manage their LTC and live a fulfilling life alongside it can provide a sense of hope to CYP. Their interaction with supportive others within an intervention that considers their LTC within the context of their daily lives appears to reinforce to CYP that they are more than just their illness and that they too have something to contribute within their relationships with peers and intervention deliverers. This sense of mutuality can also contribute towards developing feelings of empowerment. Having a sense of hope may provide the

motivation for CYP to engage with an intervention and implement the knowledge that they have learned, whilst increased resilience may further contribute to their sense of hope for the future.

indicates a cyclical relationship between the themes and suggests that the impact of an intervention may affect future engagement. One of the key factors which may be associated with maintaining engagement over time is the ability of the intervention to flexibly adapt to the physical and mental health needs of the CYP for whom it is intended. This reflects an important point raised by our study-specific CYP Public and Patient Involvement group who were consulted on emerging preliminary overarching themes identified by this review. They emphasised that they preferred it when individuals delivering the intervention listened to what the young people themselves felt was the main issue, rather than trying to impose an intervention they felt was irrelevant upon them. There is a tension between the need for an intervention to be flexible and provide a sense of being unconstrained, and the need for boundaries. Evidence suggested that whilst CYP welcomed the opportunity to escape the worries and constraints associated with living with an LTC in everyday life, they required certain limits on behaviour, discussion topics and physical activity in order to experience a space as safe and appeared to be reinforced by both intervention deliverers and peers. The apparent balance required to ensure these two concepts are incorporated into an intervention may represent a challenge to those who design and deliver it.

Discussion

Summary of findings

Within the sixty articles included in this review, 21 types of intervention were evaluated. These included online interventions, camps, music therapy, psychoeducation and development of coping skills. The LTCs seen most frequently within the studies were cancer (14studies), a mix (10 studies), HIV (8 Studies) and T1DM (6 studies). Whilst no formal mental-health diagnosis was required for inclusion in the review, reported mental health and wellbeing aims of interventions included goals to promote adjustment, reduce anxiety and depression and improve coping skills. Articles included were typically of good study quality; the general

weaknesses in this literature included a failure to make the theoretical perspective of the author explicit, to adequately describe the context or setting of qualitative research and lack of clarity in the description of the intervention.

Results in context

Relation to previous reviews

This review is the first to examine the experiences of participants regarding interventions aiming to improve the mental health of CYP with LTCs. The majority of studies (52/57) evaluated interventions which aimed to improve others aspects of CYP functioning. This synthesis has brought together previously suggested theoretical understandings of the mechanisms underlying how the development and prevention of mental health in CYP with LTCs and expanded on them.

Wallander and colleagues' Disability-stress coping model of adjustment to chronic illness (See *Chapter 1*) acknowledges the importance of involving the family and social support under social-ecological factors that may impact on the coping strategies used to moderate the stress of having an LTC. They also discuss the importance of "perceived competence", "effectance motivation" ("a desire for understanding, predictability, and control over one's environment"[227] p. 412), and "problem solving ability" in how an individual appraises a stressor and the type of coping strategy they use to manage it. These concepts appear to be similar to the themes of 'Empowerment' and 'Skills and Knowledge' as discussed above. The line of argument presented in shares specific resistance factors within Wallander and colleagues' model which may affect adjustment to life with an LTC.[36]

The concept of social support was common within the studies included in this review and as could be expected, was mentioned most often with regard to interventions that encouraged interactions with peers and family members. Stewart and colleagues[183, 222] proposes that social support can be thought of as consisting of several components, including; affirmational, emotional and informational support. Gaysynsky and colleagues[216] go further and draws upon the typology of social support proposed by Cutrona and Suhr,[228] by suggesting that there are multiple types of social support; including Encouragement, Esteem, Network support and Tangible Assistance. The content of esteem support encompasses ideas such as;

relief of blame, validation and compliment, which could be considered as being related to the 'Self-Esteem' theme under the construct 'Resilience' from the synthesis of this review.

Venning and colleagues[44] conducted a systematic review that focused on young people's experiences of chronic illness. Their objective was to make recommendations regarding ways mental health difficulties could be prevented within this population. Their results link closely with the results of the synthesis in this review in several areas. Firstly, within Venning and colleagues' review,[44] CYP discussed how an LTC made them feel different and that other people did not understand them. This relates to the themes 'I Am not Alone' and 'Therapeutic Relationships' within the current review, which explore how much CYP valued feelings of connectedness and belonging with similar peers and empathy/validation from intervention deliverers. The themes 'Unconstrained' and 'A New Normal' within this review appears to be supported by Venning and colleagues' findings that CYP viewed the LTC as something with restricts participating in "normal" life and everyday activities and is a source of uncertainty both in the present and with regard to worries about their future.[44] The importance of social support, development of coping strategies and a sense of hope and acceptance are also discussed in both reviews. The synthesis in this review evaluates whether existing interventions are perceived to meet the needs identified by Venning and colleagues across a broader range of LTCs and identifies additional themes, such as the role of 'Hope and Inspiration' for health and social care professionals to consider when designing an intervention.

Relation to the UK health setting

Mental and physical health services for CYP within England are often commissioned and delivered separately.[229] Whilst some integration of psychological services into physical health wards for CYP with LTCs does exist, the extent to which this takes place is influenced by available funding and perceived clinical need/importance/priority and thus subject to regional variation. In 2012/13, only six per-cent of NHS mental health funding was allocated to services for CYP[230] and between 2013/14 and 2014/15, services providing mental health and social care services to CYP have seen cuts to funding of £35 million.[231] Thus recommendations made by this review regarding the need for easily accessible, evidence-based interventions that are sustainable with limited resources over time, with an impact which can be sustained beyond the end of an intervention appearing particularly relevant

The difficulty in funding and accessing holistic services may make it more challenging for mental health service providers to meet some of the conditions recommended by the "availability" "safe space" and "more than just my illness" themes. Opportunities to meet other peers with LTCs outside of physical health wards can vary according to different service providers. This may limit opportunities to for CYP with an LTC to access the social support offered by similar peers and, in turn, develop a sense of hope and resilience. The finding that MH interventions can be delivered effectively by individuals without specialist MH training, across a variety of settings, could overcome this limitation. Proposals to increase the MH support for CYP in schools and other healthcare settings offer the potential for the delivery of MH interventions to be more flexible, with more opportunities for CYP with an LTC to meet.[229] However, the issue of ensuring adequate resources are available to provide the training and ongoing supervision to ensure the appropriate, sustainable delivery of these services remains

Interventions within online and/or group settings may be another way of meeting the mental health needs of CYP with LTCs, however these approaches will not be suitable or acceptable to all CYP. Care is needed to ensure that alternative options are available when appropriate and that the design of an intervention can be adapted to meet the needs of individual members. A potential challenge for health and social care services will be on how to ensure that an intervention space is experienced as safe and allows the development of empathetic, supportive relationships with peers and intervention deliverers to develop over time within a system where waiting times for routine appointments within Child and Adolescent Mental Health Services reached 32 weeks in 2015/16.[232]

The inclusion of nine American camp interventions also raises the issue of how transferable some of the themes are to UK health and social care settings. This is particularly relevant for the themes 'Safe Space' and 'Unconstrained'. Camps could be considered as a respite intervention,[190] which is a familiar concept within UK services supporting individuals with learning disabilities and can include the provision of holidays for carers and/or those they care for.[233] This suggests that there may be a potential need to evaluate the psycho-social benefits of respite opportunities for CYP with LTCs in the UK.

The strengths and limitations of the review, and the implications and recommendations for future work are discussed in Chapter 6.

Chapter 4 Overarching synthesis

> *An integration of insights generated via studies addressing different types of questions contributes to a more synergistic, holistic utilization of research evidence.*([234]p. 89-90)

Aims

The aim of this overarching synthesis was to draw together the findings from the previously reported systematic reviews.

We used a mixed methods synthesis approach to analyse the evidence from Review 1 and Review 2 together. While all studies included in Review 1 involved CYP with elevated symptoms of mental ill health, the majority of studies in Review 2 did not assess the mental health of participants. Thus the degree to which CYP with elevated mental ill health were represented in the studies included within Review 2 cannot be determined. Based upon the author-reported mental health aims of the studies, the mental health aims of interventions tested in Review 1 more often sought to improve symptoms of mental health disorders, whereas in Review 2 interventions more often aimed to benefit broader aspects of mental health and wellbeing, such as coping and self-esteem, and reducing stress.

Although different intervention types appear in the two systematic reviews, the factors that may enhance, or hinder, the effectiveness of interventions and / or the successful implementation of interventions suggested by Review 2 can be used to help interpret some of the Review 1 findings. Likewise Review 1 evidence can help verify or refute ideas about beneficial intervention components proposed by Review 2.

Background

There are few established methods for the integration of findings from quantitative and qualitative systematic reviews of interventions. One mixed methods approach to conducting systematic reviews involves the completion of comprehensive syntheses of two or more types of data (e.g. quantitative and qualitative) before these reviews are aggregated into a final, combined synthesis. Sandelowski and colleagues refer to this framework for conducting mixed methods systematic reviews as 'segregated'.[235] Published examples of syntheses that combine separate quantitative and qualitative systematic reviews tend to take the findings of one of the reviews as their start point.[236-238] For instance, Thomas and colleagues [239] identified participant views in their qualitative synthesis of barriers and facilitators of fruit and vegetable eating in children.[239] These qualitative recommendations formed a framework for analysing the extent to which findings from controlled trials were in agreement.

Due to the differences in research questions and interventions that were included in each of the systematic reviews in this project, we felt that it would be limiting to use a model where one review was used to explain the findings from the other review only. We therefore used a similar method to the initial step used by Richardson and colleagues in their overarching synthesis of ADHD interventions in school settings to consider the findings of each review in light of the other. [106]

Method

We conducted the overarching synthesis by undertaking a collaborative question and answer exercise with preliminary findings from each review. This allowed for the issues raised during the question and answer exercise to also contribute to the synthesis of the individual reviews (see *Figure 24*). This was a deductive approach as questions based on the findings of each review were generated and used to interrogate the other review for information that could potentially inform the findings or explain gaps in the literature. Questions were framed using the format 'Review 1 found X, can Review 2 inform these findings?' Questions were related to either the synthesised review findings or descriptive details regarding included studies.

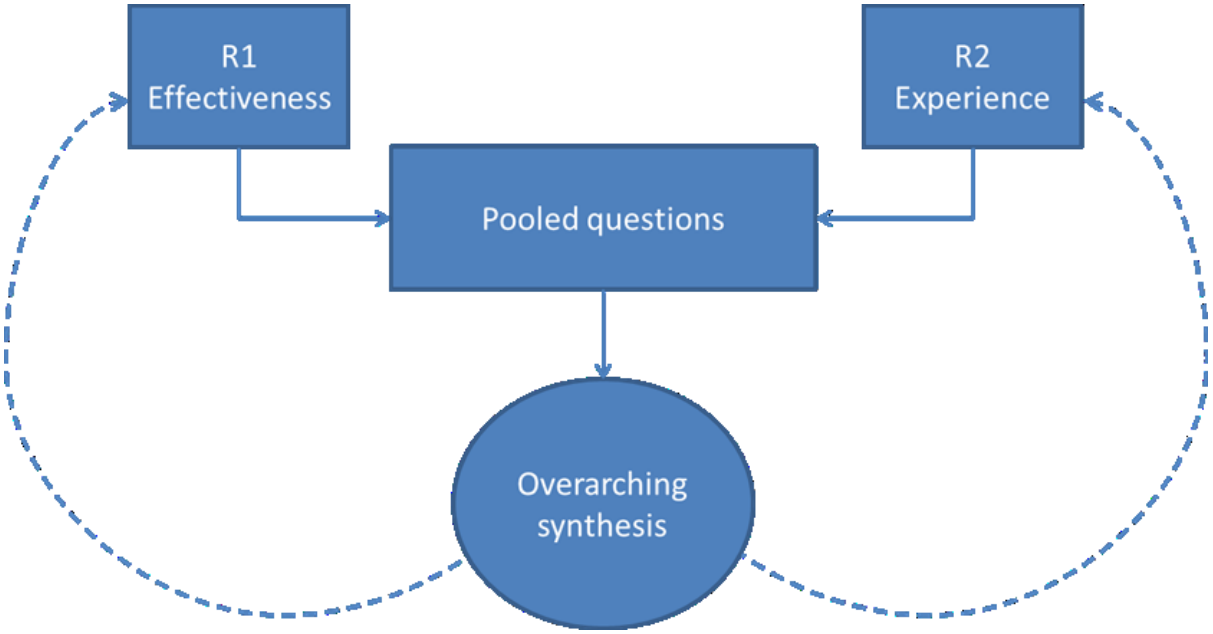

Figure 24: The overarching synthesis process

MN and LS wrote questions arising from Review 1 and Review 2 respectively. MN answered questions posed to Review 1 in written form and DM answered questions from the perspective of Review 2. The initial drafts of these questions and answers were shared with the other reviewers (MN, LS, DM) for comments and edits. In answering the questions, reviewers consulted both the systematic review findings and the data extraction forms from included studies. The resultant sets of questions and answers were grouped into categories according to shared ideas within the questions and/or answers. Each category was written up in narrative form.

Findings

Nine categories emerged from the analysis. Descriptions of the categories, the contribution of each systematic review and the implications are tabulated later in this chapter.

Degree of Overlap Between the Two Reviews

Because of a difference in inclusion criteria and search terms in Review 1 and Review 2, one would have predicted differences in terms of populations and interventions included. The requirement for elevated symptoms of mental ill health in Review 1 only, indicates that samples for Review 1 are likely to have worse symptoms of mental ill health than samples for Review 2, where this information was unknown in all but one study that did assess mental

health at baseline.[118] The database search for Review 2 included broader mental health and wellbeing terms to locate literature that may have focused on participant views rather than intervention aims in abstracts. Therefore a broader range of mental health intervention aims may have been seen in Review 2, although Review 1 included interventions with broader mental health aims that included stress, adjustment, coping and wellbeing.[119, 121, 122, 129]

Specific combinations of intervention types and LTCs were rarely seen in both Review 1 and Review 2 (See *Supplementary Materials 3* – intervention and LTC map). The only time this occurred was when the same researchers conducted both quantitative and qualitative evaluations of interventions.[118, 129, 189] Otherwise, cancer and music therapy was the only LTC-intervention dyad seen in both reviews, with Bufalini (2009) conducting a RCT and six qualitative studies exploring experiences of music therapy for CYP with cancer.[205, 207-211] This meant that the available evidence often prevented direct comparison of quantitative and qualitative research findings across similar interventions and populations.

There were also noticeable differences in the type of interventions studied in Review 1 and Review 2. CBT was the most frequently studied intervention in Review 1, with seven included studies,[82, 83, 123, 125, 129, 145] whereas only four studies featured interventions that included an element of CBT in Review 2[187, 189, 191, 198] and these were commonly combined with other intervention approaches including psychoeducation and physical activity. Parenting interventions[117, 127, 130] and group play therapy[132, 134, 137] were each the subject of three included studies in Review 1 and no studies in Review 2, Conversely, the most common intervention categories in Review 2 were online support interventions targeting various mental health and wellbeing constructs (n = 13 [182, 183, 188, 197, 202, 214, 216, 217, 219, 221-225]), Camp (n = 9 [168, 170-172, 174, 178, 179, 184, 190]), music therapy (n = 6 [205, 207-211]), face-to-face peer support (n = 4 [167, 180, 181, 218]), psychoeducation (n = 3 [176, 198, 220]) and coping skills (n = 3 [189, 196, 202]).

There were more similarities between the two reviews in terms of the populations studied. In both reviews CYP with cancer was the most commonly studied population (five studies in Review 1 [122, 132, 134-136] and 14 studies in Review 2 [178, 196, 199, 205-213, 217]). CYP with T1DM were included in four Review 1 studies [117, 119, 121, 129] and also frequently appeared in Review 2 (n = 5 [167, 189, 195, 202, 219]) and asthma appeared in three Review 1 studies [118, 137, 138] and five Review 2 studies.[118, 183, 197, 220, 223] There were, however, a larger number of studies in Review 2 that included CYP with HIV+ (n = 8 [173, 177, 179, 181, 192, 201, 215, 216]) and more than one

LTC (n = 10 [169, 172, 180, 182, 183, 193, 197, 213, 214, 223]). Only one study in Review 1 included CYP who are HIV+[120] and no Review 1 studies included more than one LTC.

What is noticeable from the map of interventions and LTCs studied across reviews (see *Supplementary Materials 3*) are the pockets of research that have been conducted for certain intervention-LTC dyads. For instance, qualitative studies of camp interventions are most often focused on cancer[168, 178, 179] and heart disease.[170, 171, 174, 190] For Review 1 three studies focus on CBT for IBD, although this may be partially explained by shared authorship across the three studies.[83, 124, 125] In Review 1 both trials evaluating stress management do so in samples with T1DM.[119, 121] It is unclear why there is a focus on particular combinations of intervention and LTC in these instances. On the other hand, online support group interventions, appearing frequently in Review 2, were evaluated across populations with ten different LTCs. This may indicate that online support groups target aspects of mental health which are common across populations. Clearer reporting of the rationale for interventions, as well as proposed mechanisms of effect, may help explain why certain interventions are used with particular LTCs.

Availability of Up-to-date, Good Quality Research

Seventy-six per cent of studies included in Review 1 and 82% in Review 2 were initially published after 2008, and 36% in Review 1 and 26% in Review 2 since 2014. Despite the relatively recent body of research, in approximately a third of the studies (n = 9) included in Review 1 there were multiple areas for concern in terms of study quality. In particular assessment of adherence, compliance or fidelity, explaining missing data, blinding of assessors and inclusion of a follow-up after treatment were identified as areas of weakness. In Review 2, there were also a number of areas of concern with the quality of the qualitative articles, including adequate description of the theoretical or ideological perspective, the context or setting of qualitative research and the clarity of intervention description. Our findings in terms of the quantity and quality of evidence available echo those of Bennett and colleagues.[81]

What Works for Whom?

As mentioned above, there was a lack of overlap between the two reviews, meaning that it was rarely the case that qualitative research could suggest why certain interventions may or may not have been effective for specific LTC populations. However, it may be that

commonalities in intervention experience revealed across studies in Review 2 can tentatively suggest reasons for some effectiveness results in Review 1, or suggest areas of focus for future effectiveness studies.

Relaxation

Two studies found beneficial effects of relaxation for asthma, one of which included both RCT and qualitative data about their intervention which aimed to improve asthma outcomes and reduce anxiety symptoms.[118] While the relaxation techniques used in the study, including breathing retraining exercises, progressive muscle relaxation and guided imagery, were considered acceptable and feasible in asthmatic youth, and reduced anxiety compared to baseline, there was no benefit when compared to standard asthma education materials received by the comparator group. The authors suggest that this was indicative of a general lack of confidence in symptom management, and that interaction with the research team, regardless of intervention, immediately reduced anxiety symptoms. This is somewhat contradicted by interview quotes from participants within the study, which suggested they used the techniques whilst feeling angry, upset and breathless. Looking beyond this study, the 'Social Support' and 'Resilience' constructs from Review 2 suggest that CYP appreciate being able to learn techniques and strategies to manage both their LTC and their emotions, which might provide a theoretical underpinning for the effectiveness of relaxation interventions more broadly.

Music therapy for procedural anxiety

The use of music therapy in CYP with cancer to assist with their procedural anxiety featured in one study in Review 1[135] and six in study in Review 2.[205, 207-211] Studies included in Review 2 identified that CYP had positive experiences of music therapy utilised during medical treatment/procedures, indicating that music therapy may provide a distraction from the burden of having an LTC,[207, 208, 211] provide an emotional outlet and regulation tool for CYP undergoing treatment[210] and may facilitate the development of relationships between CYP and their family, health professionals and peers.[207, 208, 211] The one study in Review 1 to assess music therapy found that interacting with music (making music, playing with instruments) prior to a painful treatment procedure (such as a lumbar puncture), and listening to background music during the procedure, was effective in reducing procedural anxiety in comparison to the control group who were sedated, although this study was methodologically weak.[135]

Cognitive behavioural therapy

Synthesis of the four studies featuring components of CBT in Review 2[187, 189, 191, 198] suggests that CYP may have appreciated an approach that addressed both mental and physical health needs, as well as the behavioural components of CBT interventions as they can provide an easily understood process for CYP to follow.[187] The specific CBT intervention that was evaluated across both reviews was Best of Coping, aiming to both improve glycaemic control and psychosocial wellbeing. The RCT demonstrated that it was the least effective CBT intervention in terms of CYP mental health outcomes, providing no evidence of reductions in LTC-specific stress or improved self-efficacy compared to treatment as usual.[129] The authors commented on the poor study recruitment and retention, implying that the requirement for weekly travel was a deterrent, but also that the explicit focus of the intervention on mental health was a problem for potential participants concerned about associated stigma. Access to interventions and the potential for stigma related to intervention attendance and health needs are evidenced clearly in the Review 2 themes 'Accessibility' and 'A New Normal'.

Other physical and biofeedback interventions

Review 1 included two single studies evaluating the effectiveness of interventions in CYP with chronic fatigue syndrome and chronic pain, conditions for which there is no determined physical cause. Gordon and colleagues [128] reported no evidence that in adolescents with chronic fatigue syndrome resistance training was more effective than aerobic exercise training in reducing anxiety. Heart Rate Variability Biofeedback Therapy supplemented by at-home breathing practise had no beneficial effect on anxiety and potentially detrimental effects on depression outcomes in CYP with chronic pain.[126]

An evaluation of a combined CBT and neuro-muscular training programme in CYP with fibromyalgia, a diagnosis associated with similar physical symptoms of uncertain origin, was included in Review 2.[191] The intervention was well received by participants, who valued the opportunity to address both the physical and cognitive factors which impact on their LTC, in addition to gradually increasing their physical exercise alongside a group of similar peers. The interventions studied by Gordon and colleagues[128] and Yetwin[126] appear not to have included both the cognitive and social components addressed by the intervention evaluated by Kashikar-Zuck and colleagues.[191]

Moderators of effectiveness

Review 2 suggests that the way CYP engage with an intervention is affected by age, for instance Nicholas and colleagues noted that younger and older children had different preferences regarding an intervention that included online games, education and a peer support network.[182] However, no Review 1 studies investigated differences in effectiveness outcomes by variables such as age. There were also too few studies investigating similar populations and interventions in Review 1 to allow moderator analyses at the review level. In Review 2, eight studies assessed the effectiveness of the intervention, although seven of these were not RCTs and therefore were excluded from Review 1.[118, 195, 198, 199, 201, 205, 215, 219] But, aside from Bignall and colleagues discussed above,[118] there was no clear indication from any of these studies why interventions may or may not have been effective and how individuals' experiences differed.

Category summary

Overall, due to the lack of cross-over between the types of intervention and LTC populations included in Review 1 and Review 2, the ability to explain what works and for whom is limited. Future research needs to include qualitative process evaluations alongside RCTs and vice versa to investigate reasons for effectiveness and any individual differences.

Adaptations to Interventions and Flexibility

This category considers evidence from both reviews pertaining to whether and how interventions are adapted to respond to participants' LTC or other needs. In Review 1 the majority of the interventions assessed were not described as including content adapted for the LTC of samples (see *Appendix 5*). However, six out of seven CBT studies included intervention content that was adapted to respond to the LTC of recipients. The clearest example of flexible delivery in Review 1 was for Szigethy and colleagues, where the PASCET-PI intervention was simplified for younger participants.[125]

Review 2 suggests the need for flexibility in intervention content, delivery or structure in order that CYP with LTCs can access and engage with interventions. There was evidence for a wider range of adaptations to make sure interventions were accessible to individuals including changes according to age, development and education levels, as well as LTC. There is the issue of how to tailor programmes without loss of fidelity to the intervention. In Review 1 the fidelity of, or adherence to, adapted CBT interventions was measured in several studies,

including the one study that delivered the intervention flexibly according to participant age.[83, 124, 125] There is a balance between ensuring that the effective components of an intervention are delivered as intended and ensuring that the individual needs of those receiving the intervention and the context in which the intervention is being delivered are acknowledged.[240]

Examples of the adaptation of CBT in Review 1 include the identification of LTC-specific stressors and developing coping strategies that adapt to IBD symptoms,[124] addressing fears specifically related to physical pain[123] and integrating illness narrative techniques and emphasising healthy IBD-related cognitions and behaviours.[83, 125] CBT interventions for IBD and functional somatic complaints (TAPS,[123] TAPS+IBD,[124] PASCET-PI,[83, 125]) showed evidence of some beneficial effect on CYP mental health outcomes. However, one 'cognitive behaviour intervention' (CBI[82]) showed large improvements in depression outcomes for CYP with epilepsy who were at risk of depression, and this intervention was not adapted to the LTC.

The Best of Coping (BOC) CBT intervention assessed in the study by Serlachius and colleagues[129] began as a generic CBT intervention aimed at improving coping and reducing stress.[241] Following patient feedback analysed in a qualitative study included in Review 2,[189] a number of diabetes-specific components were integrated into the intervention, including diabetes-specific goal setting and self-efficacy. Despite these components, Serlachius and colleagues[129] reported no improvement across any outcome for their intent-to-treat analysis as reported in Review 1. But a 'per protocol' analysis suggests a significant reduction in stress at the 3-month assessment for those who attended sessions, compared to the control group.

A tenet of the Review 2 'Getting In and Staying In' construct is that interventions need to be relevant to young people and, if applicable, their families. A key component of interventions being considered applicable and engaging was perceived to be the ability to use the interventions flexibly in accordance with the LTC. Within a camp [172] and online intervention [182] which were open to CYP with different diagnoses and ages, CYP were able to choose what activities they took part in in order to meet their own psycho-social needs at developmentally appropriate level. Review 2 highlights the need to be flexible according to more than just the LTC of CYP. Factors to consider also include participant age and developmental level,[169, 173, 176, 179, 182, 187, 189, 192, 196, 202, 203, 206, 207] and personal interests.[167, 177, 182, 187, 196, 207, 212]

When considering whether an intervention meets certain recipient needs, it should be acknowledged that these needs may change over time, as alluded to in several studies from Review 2.[176, 184, 194, 206, 208, 211] Developmental, educational and LTC changes could affect participation in, or effectiveness of, an intervention. A number of Review 1 studies cite a wide variety reasons for dropping out of programmes such as parental break-ups,[127] being too busy,[136] moving from the local area,[83] worsening symptoms[132] or simply lack of interest.[83] Whilst there may not be a solution to all of these potential challenges, interventions which are more adaptable to the situation of the recipients and can be adapted accordingly, may benefit from greater retention over time.

Further evidence is needed to understand whether and how interventions should be adapted for the LTC of CYP and the extent to which interventions can be used flexibly without compromising fidelity. When aiming to meet the needs of the intervention's recipient, it is important to consider how these may change over time.

Accessibility and Delivery of Interventions

Setting

Review 2 findings suggest that participation can be facilitated by utilising families' existing links with health and social care professionals. There was some evidence that future accessibility had sometimes been considered during interventions developed for research included in the reviews. For the intervention targeting anxiety in CYP with IBD as tested by Reigada and colleagues,[124] part of the rationale was to increase accessibility to mental health treatment in clinical health settings. Review 2 provides evidence that CYP and their families appreciate the delivery of mental health interventions in familiar settings, inclusive in terms of travel and any other costs,[118, 177, 180, 181, 183, 187, 188, 191, 192, 207] where they could access multiple services relevant to their needs.[180, 181]

Evidence from both reviews suggest that in addition to the physical location of the intervention, the familiarity of the environment[135, 180] and staff to CYP[204] and the extent to which it creates a therapeutic atmosphere may influence the extent to which it is perceived as accessible and may thus affect the degree to which CYP and their families engage with the intervention. There is little evidence from Review 1 that familiarity was taken into consideration when delivering interventions, other than in Bufalini's study,[135] where the child

was shown around the room where their painful procedure would take place. A number of Review 1 interventions may have encouraged practice in convenient settings, for instance home[118, 138] school,[118, 131] or delivered over the phone.[83, 125, 127]

Role of technology

Whilst it was not possible to determine whether interventions delivered online or by telephone are as effective as those delivered in face-to-face settings in Review 1, the review does provide some evidence that interventions delivered, at least in part, over the phone can be effective in relieving mental health difficulties.[83, 125 195]

Results from Review 2 suggest that some CYP and parents appreciate online interventions which CYP can access with minimal support from parents and the freedom of expression which can be facilitated by perceived anonymity.[182, 183] However, other studies suggest that despite the convenience of online interventions, CYP and their families may still prefer face-to-face meetings.[194] Some staff noted that for interventions delivered online it was harder to monitor how the intervention was received due to being unable to read the body language of the participants.[183] It was also noted that whilst some participants actively engaged with online interventions, it could be difficult for shy children to join in initially;[183] whilst others only participated when they had a specific problem.[188] Studies within Review 2 also noted the importance of a facilitator in encouraging initial engagement and discussion, particularly in the early stages of an online intervention.[182, 183, 202, 217] Review 1 does not include any studies that take advantage of technologies CYP use in everyday life.

Delivery

The 'Unconstrained' theme within Review 2 suggests that CYP value opportunities to feel that they can "escape" from worries and everyday routines associated with their LTC, including hospital visits.[171, 172, 192, 207] Interventions which allowed CYP to try fun activities, or enabled them to "be themselves"[172](p. 115) with similar peers, were also highly valued. In Review 1, group play therapy interventions promoted interaction with peers in similar situations and potentially elements of 'fun'.[132, 134, 137]

It is not possible to determine whether there were any differences in effectiveness according to individual's specialist training, as there were too many confounding variables. Review 2 suggests that some interventions delivered by individuals without specialist training,

such as peer-mentors, were acceptable to CYP and in some cases were perceived as being more approachable or trustworthy when based upon the deliverer's own experience.[179] Evidence from the 'Keeping It Going' theme suggests that adequate training is required to ensure that both specialist and non-specialist individuals deliver the intervention in the way that it is intended and that the benefit of the intervention is sustained over time.

Stress and Coping

The following section considers concepts that emerge from the two reviews that relate to managing the stress of living with an LTC, and evidence for strategies to help CYP cope.

Managing physical symptoms alongside daily life

The 'Hope and Inspiration' and 'A New Normal' themes within Review 2 indicate that having one or several role-models who are living successfully with a LTC and being able to incorporate the realities of living with an LTC into everyday life as well as taking part in "normal" activities, can provide CYP with a sense of hope.

Eleven interventions from Review 2 included components that focused on supporting CYP to improve control of LTC symptoms, while the effect of interventions on LTC symptoms was measured in 13 of 25 Review 1 studies. The 'Resilience' construct from Review 2 described how CYP perceive learning skills which would allow them to take responsibility for managing their LTC, improve their confidence and empower them to move forward with their lives.[187, 191] There is evidence from Review 1 that where interventions successfully supported CYP to manage physical health symptoms, there was also a beneficial impact on their mental health,[123, 125, 138] although this was not always the case.[118]

Choice, mastery and having a voice

Review 2 suggests that interventions which support CYP to make choices can facilitate feelings of empowerment. Despite the inclusion of two palliative care interventions which encouraged CYP to make decisions about their care,[136, 140] it was not possible to assess the extent to which the availability of choice as a component of the intervention was beneficial or not.

Review 2 indicated that CYP reported benefits in terms of empowerment and self-esteem from opportunities to experience mastery and personal autonomy. These experiences were not

restricted to management of LTC, but were also achieved through taking part in fun or challenging activities and having the opportunity to reflect on their illness journey.[184, 206, 207] None of the studies within Review 1 explicitly evaluated the impact of interventions on self-esteem outcomes, although developing a sense of 'positive self' and LTC-specific self-esteem were components of some CBT interventions.[83, 129]

Review 2 also suggests that interventions that encourage children to have a voice within the intervention were important components of empowerment. Studies within Review 1 which encouraged CYP to voice their opinions included two palliative care interventions, which asked children about their wishes[136] and included CYP in advanced decision making for end of life care.[140] While several Review 1 interventions incorporated aspects of problem-solving or goal-setting, it was often unclear to what extent this was led by CYP themselves, which is something that might offer a greater sense of empowerment.[189, 194, 200] It was also unclear whether these intervention components had an influence on outcomes.

Distraction

Both reviews offer some support for the use of distraction as an effective intervention when CYP are undergoing distressing procedures. From Review 1, Bufalini reported large beneficial effects of music therapy as a distraction during a painful procedure, although the study was at high risk of bias.[135] Likewise, Review 2 indicated that interventions such as music therapy and making videos were perceived to reduce anxiety.[207, 211, 212] Distraction alleviated the boredom associated with long hospital stays.[182] Distraction was also seen outside of the context of procedural distress. In Review 1 Szigethy and colleagues included distraction in an attempt to avoid negative cognitions.[83] However, Review 2 suggests that being able to tolerate some distress is important for CYP to access and maintain engagement with an intervention.[176]

Coping over time

To sustain the impact of interventions on CYP wellbeing over time, the 'Keeping It Going' theme within Review 2 proposed that interventions should incorporate the use of new skills into the daily routine. There is evidence of interventions across both reviews which encourage this through homework tasks for parents to practice new skills and manage CYP behavioural problems[127, 130] and CYP to practise exercises, relaxation, CBT techniques and stress management skills between sessions.[118, 126, 128, 138, 191, 195, 200, 202]

There are a number of proposed methods for reducing stress and improving coping in the short and long term that arise from the two reviews. While having a role model who represents an achievable, 'normal' life appears to be valued by CYP, none of the Review 1 studies incorporated this into their interventions. Encouraging active decision making, autonomy and participation in discussions about the child's own illness is one strategy which may lead to increased self-esteem, Review 1 was unable to effectively evaluate these suggestions.

Working with Family and Peers

This category explores the importance of involving others in mental health interventions for CYP with LTCs including interventions which involve members of the CYPs family, or members of their community such as teachers, are explored. Studies included in Review 2 indicated that interventions that aim to treat the mental health of CYP were perceived to improve the quality of family relationships[169] and provided families with the opportunity of meeting others in similar situations, which may provide a sense of community and support.[169, 176, 190] Parents stated that it was sometimes challenging to answer their children's questions when they did not know the answer[176] and that they appreciated interventions that enabled them to support their child.[196] Some interventions appeared to enable parents to cope with daily stressors and adopt a more positive perspective with regard to the family's situation/health of ill child.[169, 171, 182, 189, 192]

Review 1 suggests that parenting interventions may help improve parents' feeling of parenting competence and confidence, as well as improving their own mental health.[117, 127, 130] Review 2 also provides some evidence to suggest that skills learned by families may in turn positively impact on the mental health of their child by promoting positive coping strategies within the family.[190, 194, 196]

The potential for tension between providing the opportunity for CYP to become empowered via learning to manage their LTC versus involvement of parents in the intervention process is acknowledged. Parents have an important role in supporting CYP to manage their LTC, attend interventions and practise techniques which may benefit their mental and physical health; as acknowledged within the 'Getting In and Staying In' construct within Review 2. However, some studies in Review 2 also acknowledge how parents value the opportunity to

step back and how CYP may benefit from accessing "adult free" spaces [174](p. 284).[185, 190] In Review 1, five interventions had components that were delivered to parents as well as CYP,[83, 124, 125, 140, 145] although only Lyon and colleagues[140] intervened in a way that simultaneously involved both CYP and their parents.

Reigada and colleagues[124] educated parents about the LTC and actively encouraged parents to foster a sense of independence in their children. The findings of Review 2 suggest that Reigada and colleagues' approach could be perceived as effective[194] and indeed, it reported large beneficial effects on LTC-specific anxiety, although this was of course a small element of the overall intervention.

There was also evidence that involving affected peers in interventions might be beneficial. Review 1 included three studies which encouraged group play or activities with affected peers.[132, 134, 137] Synthesis suggests that the group play interventions were effective, producing large beneficial effects on mental health; however, the quality of these studies was poor and no firm conclusion regarding the efficacy of studies emphasising group interaction compared with individual based delivery could be made due to study heterogeneity. However, Review 2 indicated that group interventions allowing interaction with affected peers, particularly older mentors, was highly valued by CYP due to the feelings of reduced isolation it created and access to support.[167, 171, 175, 188]

Therapeutic Relationships

Review 2 identified the theme of 'Therapeutic Relationships' as an important component for making the participants of an intervention feel as though it was a safe and secure environment, where they feel valued, respected and cared for if the relationships with fellow recipients or whomever is delivering the intervention are understanding, validating and respectful of their needs. Review 2 suggested that these relationships may take time to develop and establish trust through the exchange of personal information and may facilitate the expression of emotions. This could be a key component of a successful mental health intervention, due to its role in enabling CYP to access social support.

Some interventions in Review 1 incorporated opportunities for discussion about experiences and emotions, such as group discussions, one-to-one sessions and tasks that require participants to identify typical stressors. While this could facilitate the development of key

therapeutic relationships, there is little available evidence to examine the extent to which the sharing of experiences occurred, or whether the CYP felt that other people understood and empathised with them. The only insight available emerges from the intervention rating/satisfaction data reported in four studies.[119, 123, 124, 140] Each of these papers included findings regarding ratings of the clinician involved in delivery of the intervention. Lyon and colleagues[140] administered a satisfaction questionnaire to families receiving Advanced Care Planning (ACP), who rated all three sessions as 'worthwhile', with the vast majority of adolescents finding the sessions helpful.

CYP and parents in the study conducted by Masia Warner and colleagues[123] were highly satisfied with the therapist delivering the intervention, reporting high levels of satisfaction and treatment benefit. Boardway and colleagues[119] reported high levels of satisfaction with several components including staff, content, equipment, patient improvement and overall satisfaction, however their stress management training programme was ineffective across all outcome domains. The limited information from Review 1 studies cannot tell us whether clinical effectiveness is impacted by relationship with the therapist.

Holistic Approach

Although both reviews only included interventions that aimed to improve CYP mental health, it was clear that the majority of interventions aimed to improve other aspects of CYP's health, most often outcomes related to their LTC. In Review 1, 20 of the 25 included studies also aimed to improve other outcomes aside from those categorised as child mental health. Sixteen of these 20 interventions aimed to improve LTC symptoms as well as mental health. Despite this evidence that interventions aiming to improve mental health assessed in RCTs tend to have a more holistic focus, there was a lack of evidence of effectiveness for intervention targets beyond mental health.

In Review 2, all but five of the 57 included studies focused on interventions that also targeted outcomes other than those categorised as mental health. Frequently seen targets for interventions included social support or relationships (22 interventions), LTC symptoms (13 interventions), LTC self-management (11 interventions) or education or knowledge about the LTC (9 interventions). Review 2 intervention targets were therefore broader than Review 1.

In terms of improvement in child mental health outcomes, the interventions targeting only this domain almost exclusively reported large beneficial effects. The three group play therapy interventions,[132, 134, 137] two emotional intelligence training interventions,[131, 133] and a single music therapy intervention reported large, beneficial effects on most mental health outcomes, although these findings may be limited by the lower quality of these studies.[135] However, this raise the suggestion that focusing solely on child mental health outcomes might be an effective approach.

Table 5: Categories contributing towards overarching synthesis

Category	Summary	Review 1 contribution	Review 2 contribution	Implications
Degree of Overlap Between the Two Reviews	Limited amount of overlap between reviews. Small pockets of LTC population-intervention dyads seen.	Range of LTC and interventions studied, many gaps for specific LTC population-intervention dyads	Shared study on asthma/ relaxation. Qualitative studies on cancer and music therapy. Differences in interventions and LTCs studied.	Research needed that includes effectiveness and qualitative research for same LTC and intervention. Investigate whether certain interventions are suited to particular LTCs.
Availability of Up-to-date, Good Quality Research	Much recent research in both reviews, but lack of UK based studies for Review 1 in particular and issues regarding limited quality of research.	Many recent RCTs. No UK research. Risk of bias evident for a number of RCTs.	Majority of qualitative studies published since 2008. Some UK studies. A few issues with study quality.	Need for good quality research relevant to the UK context.

Category	Summary	Review 1 contribution	Review 2 contribution	Implications
What Works for Whom?	While Review 1 indicates some evidence of benefit for interventions in particular LTC populations, neither review can go further to explain why this may be and how intervention effectiveness may vary between individuals.	Examples of LTC population-intervention dyads where evidence for effectiveness and also where lack of evidence.	Can suggest reasons why interventions may be effective or not, but often using evidence across interventions.	Both RCTs and qualitative studies ought to explore variables that may affect whether a particular intervention is effective for individuals.
Adaptations to Interventions and Flexibility	There is some evidence from both reviews that adapting interventions to the needs of individuals, particularly their LTC, may be beneficial.	Some evidence that CBT and parenting programmes that are adapted to the needs of individuals are beneficial	Perceived effectiveness for interventions that are relevant to young people's needs and can adapt to CYP goals.	Comparison of adapted interventions versus generic interventions yet to be performed.

Category	Summary	Review 1 contribution	Review 2 contribution	Implications
Accessibility and Delivery of Interventions	Considers the role of the setting, use of technology and flexibility of an intervention in ensuring that it can be accessed by CYP with LTCs.	Some evidence that accessibility and familiarity of interventions may be beneficial, but hard to tease out from other components of interventions.	Familiar setting, use of technology and interventionists who can relate to the needs of young people all perceived to be effective.	Research that can investigate the relative importance of accessibility and how interventions are delivered versus other intervention components.
Stress and Coping	Details evidence from both reviews about elements of interventions which may support CYP to manage their LTC within their daily life, in the short and long-term.	Some effectiveness of interventions where CYP having a voice was an intervention component. A small number of interventions improve physical as well as mental health. Lack of evidence of effect for stress management training.	Adjusting to LTC key for CYP. CYP want choice and a voice. Distraction may be a useful intervention component. Different stressors experienced over time, so coping may change.	Relative importance of stress and coping with LTC highlighted by Review 2 suggests that more high quality research into interventions addressing adjustment should address this. Effective stress management is needed.

Category	Summary	Review 1 contribution	Review 2 contribution	Implications
Working with Family and Peers	Evidence from reviews that interventions ought to include family, particularly parents, and that there are benefits of group settings where CYP meet peers with LTCs.	Parenting interventions may be beneficial for parents. Unclear if parent components of CYP interventions beneficial.	Tension between involving parents and empowering CYP. Interaction with peers with LTCs was highly valued by CYP.	Further research on how to include family in interventions and whether including mentors or peers with LTC experience is a beneficial intervention component.
Therapeutic Relationships	Explores the evidence available from both reviews about the impact of CYPs relationships with their therapists, ill and healthy peers on intervention effectiveness.	Range of different interventions often incorporate opportunities for sharing experiences and emotions, although effectiveness of this component cannot be isolated.	Therapeutic relationships are an important component of interventions. Many Review 2 interventions aimed to improve social support.	Research that investigates how therapeutic relationships are established ahead before or as part of interventions is needed.

Category	Summary	Review 1 contribution	Review 2 contribution	Implications
Holistic Approach	Reviews included interventions that targeted mental health; however, the majority of interventions also targeted other outcomes.	Majority of RCTs investigated interventions that also aimed to improve LTC outcomes	Interventions often targeted social support and aspects of the LTC like knowledge and self-management.	High quality future research is needed on the broad range of interventions that might be relevant to CYP with LTCs, which should include a wider range of outcomes including both physical and psychological outcomes, rather than just diagnosed mental health conditions or aspects of mental health, but not always mental disorders.

Conclusion

This chapter drew together the evidence presented by the quantitative and qualitative systematic reviews and presented nine overarching synthesis categories which enabled a structured discussion of findings. *Table 5* above summarises the findings from the overarching synthesis and their implications. Despite the lack of good quality evidence, we are able to use the evidence presented to make a number of suggestions regarding what may impact the effectiveness of interventions aiming to improve the mental health of CYP with LTC, as well as make recommendations for further research in the field.

The main challenge for comparison in this overarching synthesis relates to the different inclusion criteria across the reviews that may have led to a focus on different interventions. Review 1 included studies where participants had elevated mental ill health scores at baseline measured using a validated and standardised scale, and therefore interventions may be more focused on improving clinical symptoms. In Review 2, the nature of qualitative studies meant that participants' symptoms of mental ill health were very rarely measured. Including any samples in Review 1 regardless of mental health measures at baseline would have significantly increased the size of the review. Future reviews might consider this and make the review manageable by focusing on particular interventions or LTCs.

It is clear from both the quantitative and qualitative evidence that interventions aiming to improve the mental health in CYP with LTCs often take a holistic approach, rarely focusing on only one aspect of mental health. Interventions in included studies often target aspects related to CYPs physical health or for the interventions featuring in the qualitative review interventions frequently aimed to improve social support. While there is not strong evidence that these interventions are effective for the range of targets, it does appear that there is a preference that interventions focus more broadly on the range of issues faced by CYP experiencing both mental and physical health difficulties. This is expressed by those involved in delivering and receiving the interventions and indicated by the aims of the interventions developed. Further research is required to investigate whether a holistic approach to interventions is beneficial. We found tentative evidence to suggest that interventions ought to be used flexibly to meet the various needs of CYP where possible, including accounting for the specific nature of their LTC. We suggest that a balance is required between the need to manualise an intervention to ensure that delivery is standardised and delivered with fidelity, versus the need for flexibility of delivery to ensure the needs of individual CYP are met. While some interventions are associated with populations with certain LTCs, for example CBT for CYP with IBD, there currently is not enough evidence to ascertain whether the effectiveness of certain interventions aiming to

improve mental health is moderated by the LTC. In order to improve the accessibility and long term effectiveness of interventions for the mental health of CYP with LTCs, Review 2 suggests future research should consider ways in which technology could improve intervention delivery.

In summary, this overarching synthesis of qualitative and quantitative evidence highlights a number of implications regarding interventions for the mental health of CYP with LTCs, although this is to some extent limited by the lack of high quality evidence. Therefore further research is warranted generally, particularly research that makes use of both high quality quantitative and qualitative evaluation in the same study and investigates further some of the messages from this synthesis.

Chapter 5 End-user involvement

Introduction

The inclusion of end-users within healthcare research is encouraged due to the valuable insight they can provide during the research process.[242, 243] By incorporating the knowledge and experiences of these groups into each stage of a project's conceptualisation, development, implementation and dissemination of findings, it ensures that the research remains relevant and accessible to the people whom it is intended to benefit.[244] Thompson-Coon and colleagues[245] conceptualise end-user involvement to include health and social care professionals who may utilise the research findings in their daily practise, in addition to the involvement of service users, carers and service user representatives typically considered in the literature as patient and public involvement.[246]

A full account of the involvement of topic experts, including psychiatrists, psychologists, paediatricians, young people and parents at each stage of this project can be found in Supplementary Material 4. In Table 6 we provide a summary of the involvement at each stage and the impact their experience and insight had on the project.

Impact of end-user involvement

On the children and young people and their parents

One of our main concerns was to ensure that as well as the research benefiting from their involvement, the CYP and their parents also benefitted. CYPAG members and parents were provided with £20 vouchers following each event to acknowledge their contribution and time commitment. We reiterated at every meeting that participants should contribute towards the project in whatever way was comfortable and meaningful for them.

Throughout the course of the project we discussed and reflected on the balance between burden and benefit. There is evidence that the young people benefitted from their involvement simply by meeting one another through this process. At the first meeting, two of the young people discovered that they shared the same rare physical health condition and each appeared to value the opportunity to meet someone else in the same situation. This impression was reinforced by email feedback received following the first CYPAG meeting from two group members:

It was good to hear other people's points of view [CYPAG member]

I think it was really helpful having other people who have gone through the same things as you that understand you [CYPAG member]

We had not intended to have a dedicated parent group providing input into the project but we observed that when parents came to collect their children from the meeting, they spontaneously discussed their experiences of accessing care for their child, issues with school attendance and their children's health conditions. Although it was not part of the original patient and public involvement plans for the project, this presented an opportunity to access the experiences and knowledge of the parents and we henceforth held separate parent group meetings in tandem with the young people's meeting. Within the parent group, parents commented how "meetings like this" [Parent, Third meeting] were good for CYP to share experiences and meet people who could "get on with their lives" [Parent, Third meeting]. One example provided was how one young person met an older peer who was in a romantic relationship, which the younger child had thought was a slim possibility for themselves, given their physical condition.

The parents agreed that sometimes it was difficult to get their children out of the house, but that they were always keen to attend these research meetings. Overall, the experience of witnessing how enthusiastic CYP were in having the opportunity to meet each other outside of a treatment setting was extremely powerful for the researchers. This observation later contributed towards the decision to have 'Social Support' as an overarching construct within Review 2, rather than a theme within 'A Therapeutic Foundation'.

On the report findings

Involvement of end-users in each stage of the systematic review process has made a valuable contribution towards the creation of a robust evidence synthesis by incorporating the experience of those whose lives the project intends to influence. As a result of this contribution, this report is grounded in the experiences of CYP with LTCs and mental health problems and the health care professionals who provide their care, and appears to be relevant to their needs. This is reflected by the degree to which the preliminary findings from Review 2 and Overarching Synthesis appeared to be transferable across different end-user groups and in keeping with practitioners' approaches to providing mental health interventions.

There was agreement across different stakeholders that the separation of mental and physical health when developing interventions was artificial and unhelpful. The views of the people

consulted have also highlighted implications for areas of future research, which are explored in the final chapter of the report.

Table 6: Description of end-user involvement event

Activity	Date; Method	Who?	End user perspectives represented	Impact
Planning stage				
Finalising search terms	January 2016; email	Co-applicants and expert advisory group (n= 16)	Paediatric psychiatry, paediatric psychology, paediatrics, patient and public involvement, parent, researchers - child mental health	Terms including 'Crohn's', 'fibromyalgia', additional cerebral palsy terms and broad mental health terms like 'psychiatric / psychological / emotional disorder' added to the search.
Defining key terms and refining inclusion criteria	11th February 2016; meeting in London	Co-applicants (n= 12)	Paediatric psychiatry, paediatric psychology, patient and public involvement, parent, researchers - child mental health & evidence synthesis	Definition of 'long term condition' finalised. Eligibility criteria for long term conditions finalised. Eligibility criteria for mental health interventions finalised.
Checking relevance of the project and definitions of key concepts	20th February 2016; meeting in London	Review team and CYPAG (n=12)	Patient and public involvement, children and young people, researchers – evidence synthesis	Reinforced the importance of acknowledging the link between physical and mental health and how interventions aimed at treating one aspect of mental health may impact on another. Strengthened our justification for including all outcomes in the synthesis of Review 1.
Review methods stage				
Study selection	14th April 2016; teleconference	Co-applicants (n= 11)		Refined our interpretation of the eligibility criteria for the reviews. By sharing some of the challenges of identifying relevant information at this early stage of the process, we were able to use the experience of the entire team in planning alternative approaches.

Categorising interventions and outcomes for Review 1; planning dissemination	10th September 2016; meeting in London	Review team and CYPAG (n=11)	Patient and public involvement, children and young people, researchers – evidence synthesis	Reinforced the importance of a wide range of outcomes and the impacts of interventions on relationships with friends and family members. CYP emphasised the importance of school attendance and coping with school, outcomes that were not frequently reported in the included studies. This discussion highlighted the importance of this type of outcome to CYP and informed our implications for future research. Decided to consider a podcast as a dissemination activity involving the CYP and to include time for editing plain language summaries for different end users in the final CYP meeting.
Discussing preliminary findings and approach to synthesis; identifying consultation opportunities	3rd October 2016; meeting in Exeter and subsequent email dialogue	Co-applicants and expert advisory group (n= 16)	Paediatric psychiatry, paediatric psychology, paediatrics, patient and public involvement, parent, researchers - child mental health and evidence synthesis	Provided a practice-based verification of our interpretation of the emerging findings. The need for caution due to small numbers of studies was emphasised. Several opportunities for consultation were identified and facilitated as a result of this meeting.
Consultation stage				
Discussing preliminary review findings; planning dissemination	28th January 2017: meeting in London	Review team, CYPAG and their parents (n=15)	Patient and public involvement, children and young people, parents, researchers – evidence synthesis	Reinforced ideas which were emerging from the analysis in Review 2, such as the importance of having a voice within the wider community. Some points which challenged the initial synthesis were also raised by members of the CYPAG. An example of this was the suggestion that mental health interventions should acknowledge that the

				needs of CYP were serious and therefore did not always need to contain an element of 'fun'. This encouraged the researchers to check the studies included within Review 2 for any refutational data. Ideas from the parent group regarding the impact of school environment, long term effectiveness of interventions and the need for an age-appropriate approach reinforced ideas presented by the CYPAG, which were drawn upon to inform the analysis for Review 2 and the Overarching Synthesis. The parent group highlighted the need for interventions to involve systems around the child and the challenges in having the mental health needs of CYP recognised and met by schools and primary care clinicians. This feedback validated the decision to keep 'Availability' as a separate theme within Review 2 and the category 'Holistic Approach' within the Overarching Synthesis. The views of parents and CYP helped researchers bring together the results from the two reviews within the Overarching Synthesis by providing an alternative viewpoint through which to conduct a secondary interpretation of their results.
Discussing preliminary findings; establishing links for dissemination	Various dates between 10th January and 15th March 2017; meetings,	Various[a]	Researchers - child mental health & evidence synthesis, patient and public involvement, children and young people, paediatric psychiatry, third sector organisations, paediatric	Highlighted methodological decisions which required additional detail within the write up of the report. Examples include, i) feedback from the Paediatric Psychiatry Liaison Group regarding the importance of considering outcome measures such as school attendance, meant that the extraction and

	Skype calls, conference presentations; teleconference (n=11)	psychology, paediatrics, parents, conference attendees at the MQ Mental Health Science meeting	synthesis of all intervention outcomes in Review 1 needed more emphasis within the written report and ii) YPMHAG members assumed there would be differences in experiences of mental health interventions across LTCs and age, highlighting that the methods section should acknowledge how the views of CYP with different LTCs were incorporated into the results. The Paediatric Liaison Group suggested some additional studies to check for eligibility for Review 1. All suggested studies had been excluded during screening as they did not meet the inclusion criteria. Consideration of the availability of evidence from samples of CYP with learning disabilities was requested by the CYPMHC. At least one young person from the YPMHAG had a precise definition of a mental health intervention as something that is used as a preventative type tool distinguished from treatment, indicating the need to define 'intervention' in the final report and particularly in the plain language summaries. Feedback through consultation events also had some influence on the synthesis for results in Review 2. Interest shown by members of the CYPMHC regarding how much the synthesis of Review 2 focused on CYP's experience of transition between child and adult mental health services led to us re-examining the data set for relevant information.

Presentation of the results of Review 1 and 2 at the MQ Science Meeting provided the opportunity to disseminate our results to an interested audience of clinicians, researchers and CYP who use mental health services. During this conference, the Mental Elf expressed interest in writing a blog post about the project, creating a further opportunity to disseminate the results in a format accessible to the wider public.

The consultation process also created opportunities for collaboration with other organisations. We shared our methods for Review 2 with the Mental Health Foundation who were undertaking a qualitative systematic review in a similar topic area. CLIC Sargent offered to share the results of several reports they had recently been working on and we reciprocated this by sharing information located through our literature searches.

Through discussing preliminary results with organisations working in the area of children and young people's mental health we were able to explore opportunities for using their communication channels as a platform for disseminating our findings. CLIC Sargent, the Mental Health Foundation, the ERG, MQ and the Paediatric Psychology Network all indicated that they would be willing to provide a platform for the dissemination of our podcasts and plain language summaries.

Dissemination stage

Creating dissemination products	18th March 2017; meeting in London	Review team, CYPAG and their parents, podcast recording specialist (n=15)	Researchers – evidence synthesis, patient and public involvement, children and young people, parents	Children and young people and their parents recorded material for two podcasts – the first discusses the findings of the project, the second their experiences of being involved. Children and young people and their parents also co-wrote plain language summaries of the findings – one for the final report and adapted versions for their respective audiences (children and young people and parents).

[a] Paula Lavis (coalition co-ordinator) and members of the Children and Young People's Mental Health Coalition, National Institute of Health Research Maudsley Biomedical Research Centre Young Persons Mental Health Advisory Group, Antonis Kousoulis and Josefien Breve from Mental Health Foundation, Helen Gravestock (Research and Policy Manager) CLIC Sargent, conference presentations at the MQ Mental Health Science Meeting, UK Paediatric-Psychiatry Liaison Group meeting, Expert Reference Group (ERG) Psychological Skills and Knowledge for Multi-Disciplinary Team Heathcare Professionals Working with Children and Young People with Physical Health Conditions, Paediatric Psychology Network Committee, paediatricians at the Royal Devon & Exeter Hospital attending their weekly meeting, meeting with a PenCLAHRC Patient Involvement Group member, meeting with a clinical academic fellow.

CYPAG – Children and Young People Advisory Group

Chapter 6 Discussion and conclusions

The aim of this research project was to evaluate the effectiveness and cost-effectiveness of interventions aiming to improve the mental health of children and young people (CYP) with long term physical health conditions (LTCs) and, to explore the factors that may enhance, or limit, the beneficial delivery of such interventions. To address these aims we conducted two systematic reviews, one focused on quantitative studies and one on the qualitative evidence related to relevant interventions. We then brought together findings from each review in an Overarching Synthesis. As part of the project we consulted with a range of end users about preliminary findings, strengthening the main arguments and developing key implications on the basis of these activities. A children and young people advisory group (CYPAG) was formed and met four times during the project to provide input into methodological decisions, consultation on preliminary findings and to help prepare dissemination materials. This final chapter summarises the findings of each review and the Overarching Synthesis, describes the strengths and limitations of the work conducted, outlining the implications for practice and recommendations for further research.

Summary of findings

Summary of Review 1

In Review 1 (Chapter 2), we synthesised 25 randomised controlled trials (RCTs) (31 articles) that assessed the effectiveness of interventions aiming to improve the mental health of CYP with LTC and elevated symptoms of mental ill health. These studies evaluated 11 types of intervention provided to CYP with 12 different types of LTC. We extracted 269 outcome measures from the 31 articles. While the primary focus of the review was the impact of interventions on the mental health of CYP; all secondary outcomes were synthesised in order to evaluate any benefits of interventions on other aspects of young people's lives, including key aspects of individual and family functioning. As many different measures of similar underlying constructs were reported, we categorised them into 28 categories. Seventeen categories related to CYP mental health, the rest related to "other outcomes".

The greatest volume of research focussed on the effectiveness of CBT, which was evaluated in seven studies (10 articles). These studies provide tentative evidence that CBT-based interventions could be beneficial for the mental health of CYP with inflammatory bowel disease (IBD), chronic pain, epilepsy and persistent functional somatic complaints, but not type 1 diabetes mellitus. The majority of the CBT interventions had content which was adapted to the needs of the LTC of CYP in the sample. In addition, four parenting programme interventions were tested in three studies. Group play therapy interventions were also assessed in three studies. Other intervention types (palliative care, relaxation, stress management training, emotional intelligence training, massage therapy, biofeedback, resistance training and music therapy) were only reported in one or two studies.

We calculated effect sizes for each study outcome wherever data allowed. Trials were typically small, so effect sizes across the included studies were characterised by wide confidence intervals. Therefore, the evidence for particular interventions used with similar samples of CYP is very limited. There were relatively few opportunities to meta-analyse studies with similar intervention, comparator, LTC and outcomes. No more than two studies shared particular combinations of these study characteristics and could be meta-analysed.

Study quality of the relatively small amount of research to date is poor, with 19 of the 25 studies having at least four separate indicators of potential bias. Blinding of assessors and evaluation of adherence, fidelity and compliance was often neglected, and reporting of randomisation methods and allocation concealment was largely unclear. Fewer than half of the studies included a follow up assessment, with only half of those occurring beyond six months post-intervention. The quality of future research in the field should therefore be improved. Although findings from Review 1 are tentative, they support previous research. A systematic review by Bennett and colleagues of psychological interventions for mental health disorders in CYP with chronic conditions also reported preliminary evidence that CBT interventions may be beneficial.[81] Review 1 is the first systematic review to consider all interventionsfor elevated symptoms of mental ill health in CYP with LTCs.

NICE guidelines recommend CBT for CYP with depression.[53] James and colleagues published a Cochrane systematic review of CBT for childhood anxiety disorders in CYP without

comorbid LTCs, reporting a suggestion that CBT is a more effective treatment than treatment as usual or active controls.[69] Given our own findings, there is tentative evidence that these recommendations may hold for CYP with LTCs. Review 1 does not explain why CBT has promising effects, or whether and how to adapt it to CYP with LTCs for the best effect. Future research should seek to explore these questions.

The findings of Review 1 point to the need for large, high quality RCTs with consistency in intervention, design and outcome reporting. Rigorous testing of interventions across a range of conditions and locations, including fidelity and process evaluation measures would aid comparison of the components of different interventions. It would be particularly useful to examine whether intervention types that show some promise (e.g. CBT) are effective across a range of LTCs, including whether (and how) they should be adapted to specific LTCs. Although we included studies of interventions that aimed to improve mental health, the majority also targeted other outcomes, particularly LTC symptoms. Therefore, future research might consider treatment integrating both physical and mental health care, and its effects across a wider range of outcomes, rather than focus primarily on mental health in terms of population, intervention and outcome.

Summary of Review 2

In Review 2 (Chapter 3), 57 studies were reviewed, which included qualitative data collection and analysis on the experience of, and attitude towards, interventions aiming to improve the mental health and wellbeing of CYP with LTC. These studies evaluated a more diverse range (n = 21) of interventions than seen in Review 1. The most frequently seen LTC in the 57 included studies was cancer (n = 14). HIV positive intervention recipients were the focus of eight studies, while a further ten studies included CYP with a mix of LTCs, something that was not seen in Review 1. Included studies commonly explored the perceptions and experiences of interventions that aimed to improve aspects of mental health and wellbeing such as coping (n = 26), self-esteem (n = 13) and emotional support (n = 11) rather than targeting a mental health disorder e.g. depression (n = 1) or anxiety (n = 8). Interventions also often aimed to improve symptoms related to the LTC (n = 35) and social skills (n = 16). Included studies represented views from a range of different participants involved in the delivery and receipt of relevant interventions.

We used meta-ethnography to synthesise the 57 included studies (60 articles). The synthesis revealed five main constructs: 'A Therapeutic Foundation', 'Social Support', 'A Hopeful Alternative', 'Resilience' and 'Getting In and Staying In'. A number of themes contributed to each construct. We developed a line-of-argument (a model explaining how themes and constructs related to each other) that offers an explanatory model of the experience of receiving and delivering interventions to improve the mental health of CYP with LTCs (see *Chapter 3*,). The model describes the process by which CYP with LTCs access and maintain engagement with a relevant intervention aiming to improve mental health and wellbeing. Some interventions can help CYP to acquire a sense of hope for the future and increase their resilience. Interventions are often perceived to be effective when they offer participants a safe space and social support. The relationship between these constructs is not fully defined, as other factors may impact on the experience of interventions and participants attitudes.

A CYP's sense of resilience, hope and social support created through taking part in an intervention may help individuals and encourage them to maintain their engagement with the intervention. This experience may also encourage CYP and their carers to build positive relationships and use these relationships and the skills learned during interventions to develop techniques to manage emotions, physical symptoms and other relationships. Interventions that can respond flexibly to the physical and mental health needs of the CYP for whom it is intended may also help keep the CYP engaged with the intervention over time. Such flexibility, along with the acquisition of new skills, may help build and maintain the effects of interventions beyond the intervention setting. There is a tension between the perceived need for an intervention to be flexible in content and delivery to respond to individual needs and also provide a sense of being unconstrained, compared to the need for boundaries. Evidence suggests that whilst CYP welcome the opportunity to escape from the worries and constraints associated with living with a LTC in everyday life, they require certain limits on behaviour, discussion topics and physical activity in order to experience an intervention as safe. The balance required to ensure these two concepts are incorporated into an intervention may represent a challenge to those who design and deliver it.

The quality of included articles was generally good. At least ten high quality articles contributed to each theme in the synthesis. However, general weaknesses included a failure to

make the theoretical perspective of the author explicit, to adequately describe the context or setting of qualitative research, and a lack of clarity in the description of the intervention.

We believe this is the first attempt to examine the experiences of participants in interventions that aim to improve the mental health and wellbeing of CYP with LTCs. However, our synthesis shares features with some previous literature. Concepts from Wallander and colleagues'[36] disability-stress coping model of adjustment to chronic illness (e.g. problem solving and perceived competence) appear to be similar to our themes of 'Empowerment' and 'Skills and knowledge'. Some of the specific resistance factors seen within Wallander and colleagues' model, including the role of improved family relationships, sense of competence and use of coping strategies, are also seen in our line of argument.

Our themes 'I Am Not Alone' and 'Therapeutic Relationships', which explore how CYP valued feelings of connectedness and belonging with peers and empathy/validation from intervention deliverers fit with the findings of a systematic review of young people's experience of mental health in chronic illness.[44] Our themes 'Unconstrained' and 'A New Normal' are also reflected in findings from this previous review that CYP view their LTC as something which restricts participating in "normal" life and everyday activities and is a source of uncertainty both in the present and with regard to worries about their future.[44] The importance of social support, development of coping strategies and a sense of hope and acceptance are also acknowledged in both our Review 2 and the previous review.

We identified only one study in which qualitative data collection and analysis occurred alongside an RCT, which was also included in Review 1;[118] there is a need for this type of mixed methods process evaluation conducted according to best practice guidance to improve our understanding of how interventions are experienced and how this may link to effectiveness.[247]

Summary of the overarching synthesis

The overarching synthesis (Chapter 4) integrated the findings from Review 1 and Review 2. A deductive approach was used whereby questions based on the emerging findings of each review were generated and used to interrogate the other review for information that could potentially inform the findings or explain gaps in the literature. Despite the different review

questions, methods of synthesis and types of interventions seen across the two systematic reviews, the overarching synthesis allowed us to raise a number of tentative implications.

Nine categories emerged from the analysis:

- Degree of overlap between the two reviews
- Availability of up to date, good quality research
- What works for whom?
- Adaptations to interventions and flexibility
- Accessibility and delivery of interventions
- Stress and coping
- Working with family or peers
- Therapeutic relationships
- Holistic approach

There is a limited amount of overlap between the reviews, particularly when focusing on interventions used for particular populations. Much of the research included in both reviews was published in the ten years prior to the review, but there is a noticeable lack of studies conducted in the UK. While there is some evidence of benefit for interventions in CYP with particular LTC populations, for example CBT in populations with inflammatory bowel disease and music therapy for procedural distress in CYP with cancer, neither the quantitative or qualitative evidence can explain why this may be or suggest how intervention effectiveness may vary between individuals.

The review work suggests further evidence is needed to understand whether and how interventions should be adapted for the LTC of CYP. The accessibility and familiarity of an intervention's setting, use of technology and flexibility of delivery can impact perceived effectiveness and may warrant consideration as to how such issues impact effectiveness in terms of mental health outcomes and wider functioning. There was evidence from both reviews that interventions that include family, particularly parents, may be beneficial. Relationships between CYP with LTCs and their therapists, and with their ill and healthy peers seem important. While both reviews include interventions that aim to improve mental health in some way, the majority of interventions also targeted other outcomes including LTC symptoms, social support, knowledge and self-management.

Strengths and limitations

There are a number of strengths of the project's reviews, overarching synthesis and end user involvement:

- rReview 1 represents the first systematic review of RCTs evaluating interventions targeting mental health of CYP with LTCs. It was conducted and is reported in accordance with best practice guidelines.[84, 112]
- We have used comprehensive search strategies and made extensive efforts to locate research outside of academic databases.
- We have also broadened the scope of Review 1 compared to previous attempts to synthesise this evidence base by including populations with elevated symptoms of mental ill health in samples as opposed to only symptoms of mental health disorders or diagnoses.
- For Review 1, searches were not limited to English language papers, date or study location, and authors were contacted for supplementary materials and missing data where necessary. While we targeted papers reporting on interventions that primarily sought to improve CYP mental health, we included and synthesised all wider outcomes, therefore providing a fuller picture of the effects of interventions.
- To the best of our knowledge Review 2 is the first attempt to synthesise research on the experience of those involved in the delivery and receipt of interventions that aim to improve the mental health of CYP with LTCs. A strength of the review is that studies included the views of a range people with an interest in such interventions namely; CYP, caregivers and intervention deliverers on interventions at different stages in development.
- The synthesis within Review 2 considered a large number of papers in a time-effective manner and prioritised richer, interpretive findings, before using the descriptive studies to support or refute aspects of the synthesis.
- Our overarching synthesis brought together findings from both reviews. In doing so, we were able to highlight clearer implications for practice and gaps for further research.
- We consulted with a wide range of end users about our preliminary findings and used feedback to refine our analysis and write up of the final report.

- We convened a children and young people advisory group (CYPAG) that met at four points during the project. This group gave feedback on the use of terminology for the project, commented on preliminary findings, co-produced the plain English summary and recorded audio for a podcast disseminating the work.

We also acknowledge the following limitations, which predominantly stem from the primary research reviewed:

- The breadth of outcomes reported in Review 1 presented a challenge for analysis and interpretation. For example, depression was assessed using 14 different instruments across 14 different studies. The breadth of measures used made comparability across samples challenging where different scales assessing the same construct were used as the meaning of established cut points may differ across scales. Where it was not possible to find an established cut-off score in the literature for measures used, the article was considered not to have met inclusion criteria, even though it is possible the sample may have had unreported elevated symptoms.
- There were insufficient studies to allow meaningful moderator analysis of intervention type and LTC population or assess publication bias.
- No economic evaluations related to the included trials or that otherwise fit our inclusion criteria were located, which indicates a need for measures of cost-effectiveness to be incorporated in trials.
- There were no UK studies included in Review 1, therefore limiting the applicability of review findings to the UK context.
- There may be subjectivity in the categorisation of interventions. For instance, we may have grouped progressive muscle relaxation, heart-rate variability biofeedback and massage together, if there was an argument for these intervention targets or active ingredients being similar. Categorisation was on the basis of similar content and targets, meaning several interventions could not be categorised with any others.
- Each study included in this review sampled CYP participants with particular LTCs, meaning we do not have a clear sense regarding the applicability of particular interventions for improving mental health across a range of LTCs.

- Relevant interventions may have been excluded from the synthesis due to our inclusion criteria that the intervention had to be described as aiming to improve the mental health or wellbeing of CYP with an LTC.
- Due to the potential impact of misinterpretation of abstract concepts following translation from other languages, article inclusion was limited to English language article within reiview 2. There was a wide range in the type of qualitative analysis used within the studies used, which meant that the analysis conducted in individual studies was not always directly comparable. The categorisation of study findings and purposive sampling strategy as described in the methods section above meant that all studies meeting inclusion criteria were able to contribute towards the synthesis.
- It is not known how the final synthesis would have been affected if included studies had been synthesised in a different sequence. The translation component of meta-ethnography, whereby components of themes are compared to one another across articles and the line-by-line coding in Nvivo meant that the synthesis captures concepts that appear across included studies, as well as ideas which refuted the existing synthesis. Due to the wide range of interventions, utilising meta-ethnography may have meant that the constructs and themes generated from the synthesis reflect core therapeutic components common across a range of interventions. It cannot be assumed that these are applicable across all LTCs, mental health conditions or ages.
- Establishing the mental health and wellbeing focus of an intervention was not always straightforward. Only seven included studies evaluated interventions which focused only on the mental health and wellbeing of the child. In 24 studies, additional focus was on some aspect of controlling the LTC and in 22 studies on provision of social support. The small number of interventions which had a sole mental health focus may have influenced the sub/themes that emerged from the synthesis.
- None of the studies included in Review 2 sampled participants with a diagnosed mental health disorder and only two considered the current mental ill health of participants upon study entry. The number of CYP who received interventions who were experiencing poor mental health is therefore uncertain.

- Information from the quality appraisal was considered during synthesis, rather than used to exclude studies based on quality as only three articles received more negative than positive study quality ratings. At least ten of the highest quality articles (10 out of 14 "yes" ratings on quality criteria or better) contributed to each theme. This implies that lower quality articles alone did not determine any of the themes, but did provide support for them. This is common in qualitative evidence synthesis, where higher quality articles are found to contribute more to the synthesis (e.g. [160])
- The overarching synthesis was challenged by the different inclusion criteria across Review 1 and Review 2. Review 2 interventions more often focused on improving coping, stress and self-esteem than Review 1 interventions which more often aimed to improve symptoms of mental health disorders, such as anxiety and depression.

Implications for policy and practice

Given the nature of the review findings and challenges in synthesising across reviews considered in the previous chapters, recommendations for research and, in particular implications for policy and practice, can only be tentative. There is evidence that CBT may be beneficial in some of the LTC populations in which it has been evaluated (epilepsy, persistent somatic functional complaints, IBD, chronic pain). Implications for practice regarding adapting CBT for the LTC and for the effectiveness of other interventions are far less clear, although the Overarching Synthesis category 'Adapting Interventions' highlights a need for further research into any potential benefits of adapted content.

There was some evidence from Review 1 for parenting programme interventions to reduce behavioural problems in children with acquired brain injury and/or Cerebral Palsy, particularly when combined with acceptance and commitment therapy. However only four parenting interventions were included, and effects were inconsistent across studies. Three studies examining the effectiveness of group play therapy in children with cancer and asthma showed beneficial effects for anxiety, depression or coping, however these were studies of low methodological quality. Overall, the evidence from Review 1 highlights the need for further, rigorous research to be conducted, rather than recommendations for practise.

From Review 2, the 'Availability' theme indicates that it may not always be straightforward for CYP to access interventions that explicitly target their mental health. Recognition of mental health needs by physical health clinicians appears to be a particular barrier to the access of relevant interventions. Once engaged with an intervention, the importance of access was further highlighted in the 'Accessibility and Delivery' category of the Overarching Synthesis. This described the need for consideration of CYP's needs in delivery of intervention content and the possible benefit of flexible and/or technology-based content delivery. Further research on methods that might assist paediatric healthcare services to detect the mental health needs of CYP with LTCs and provide services where physical and mental health needs are managed in a genuinely integrated way, would help to plan future services to support the mental health needs of CYP with LTCs. Twenty-five of the interventions from Review 2 were delivered by someone with some degree of specialist training. Sixteen of the interventions included in this study were delivered by intervention deliverers with no specialist training. Eight of these required supervision by mental health qualified personnel and/or additional training to deliver the intervention. This indicates that it may be possible to deliver mental health and wellbeing interventions outside of mental health settings and potentially ease demand on waiting lists for mental health services. However the training and support needs of those who will be delivering an intervention would need to be considered alongside the resources required for setting up and sustaining the intervention.

Both Review 2 and the Overarching Synthesis category, 'Holistic Approach' shows that CYP value interventions that consider a range of needs (e.g. coping with school, as our CYPAG members informed us), as opposed to, for example, purely their mental health. Therefore, consideration of collaborative approaches between different treatment providers and educators may be of benefit. Research examining the extent to which collaboration already occurs in different paediatric physical and mental health-care settings could be valuable alongside an exploration of factors which may support or hinder coordinated approaches.

Evidence from Review 2 and the Overarching Synthesis also suggests that the creation of opportunities for CYP to access social support and to share feelings and experiences may result in reduced feelings of isolation and improved relationships with both peers and family members. This suggests that integration of opportunities to build supportive relationships with peers and meet

people who are successfully managing their LTC may be useful for some CYP. The constructs and themes identified in Review 2 may be considered as a framework to aid intervention development.

The method and varying locations for delivery of interventions in both reviews indicates that interventions aiming to improve mental health can be delivered outside of mental health service settings. Review 2 also highlights the importance of the involvement of family members in the engagement of CYP with an intervention and encouraging the incorporation of intervention components into daily life so that the beneficial effects are maintained over time. CYP discussed how they wanted interventions to address expectations placed upon them by wider society. This included educating teachers and friends regarding what they were physically capable of [176] in order to address the stigma surrounding certain diagnoses.[177] These findings support an important issue raised during consultation with our CYPAG, who felt that interventions should attempt to relieve the burden of feeling solely responsible for "coping" with their LTC from the child.Given the importance of social support highlighted by Review 2 and the Overarching Synthesis, relationships CYP establish with those who deliver interventions, family and peers are an important component of interventions. The relationship between CYP and provider should be considered for future evaluation in trials.

Suggested research priorities

The findings of Review 1 suggest the need for further large, high quality, well reported RCTs. In particular, improved reporting of the methods used to randomise participants and conceal their group allocation and blinding of outcome assessors is required. The production of manualised interventions would allow for more rigorous testing of interventions across a range of conditions and locations and would aid comparison of intervention components across different interventions. Likewise in order to evaluate the beneficial components of interventions across trials, better description of the content of interventions is necessary. Adherence to the many available design and reporting guidelines would facilitate this (e.g.[248]). UK-based research is also crucial to ensure that the findings are relevant to the UK healthcare system and UK CYP.

Combining the collection of effectiveness research with that of the views and experiences of those giving and receiving the intervention within the same study is necessary to explore why

certain interventions may (or may not) be effective and to identify the moderators of effectiveness. Mixed methods process evaluations conducted according to best practice guidance are required in this field as none were located as part of RCTs. Future intervention trials should clearly specify the aims of interventions, primary outcomes and preferably include logic models to demonstrate the theoretical framework of the intervention.[249] This work should also include analysis regarding the impact of different moderators, such as age, ethnicity, mental health difficulty and LTC on intervention effectiveness.

Review 1 raises implications for a research programme where an intervention with promise, such as CBT, is delivered across a range of LTCs, including variation of how the intervention is adapted to specific LTCs versus the recommended treatment for mental ill health. Such research would still need to take account of the recommendations above.

Stress and coping skills are recognised as important in both reviews, which indicates that the focus of interventions should not only be on mental health disorders and their symptoms. Future research might focus on integrated treatment targeting mental and physical health and its effects across a wide range of outcomes for those with an LTC, rather than focus primarily on mental health in terms of population, intervention and outcome.

In order to improve the accessibility and long term effectiveness of interventions for the mental health of CYP with LTCs, Review 2 and the Overarching Synthesis suggests future research should consider ways in which technology could improve intervention delivery. For example, the use of the telephone or internet to deliver therapeutic content or keep appointments with clinicians could be beneficial, while top-up or booster sessions, including those giving skills to parents, might help in sustaining intervention effects. Meanwhile, built-in flexibility to adapt to the changing developmental or LTC needs of the patient is important. Further research that evaluates the long term effects of interventions that meet these needs is required.

Review 2 in particular notes the part that family, peers (both ill and healthy) and practitioners can play in the experience of interventions, in terms of relationships with CYP or as fellow intervention recipients. Further research should consider this either as part of interventions being tested or by investigating the impact of actively involving peers in interventions.

Consensus on the use of outcome measures should also be considered a priority.[250] Having agreed gold standard outcome measures for particular mental health constructs in specific situations would further aid the production of comparable data across research outputs, strengthening future evidence synthesis. The lack of identification of primary and secondary outcomes in many RCTs, along with the large numbers of measures typically used per study raises the question of the extent to which interventions were targeting different aspects of mental health compared to wider individual and family functioning. Future trials should clearly indicate primary and secondary outcomes.

Some outcomes (e.g. LTC knowledge, self-management, school attendance, goal-setting, self-harm, hope) were identified as important in Review 2 and in our consultation with end users, but were not commonly assessed in the effectiveness studies. Consideration should be given to the best way to measure and incorporate these into future trials. Patient and other end user involvement in trials may assist the selection of outcomes that are pertinent for those involved.

No relevant cost-effectiveness studies were located by Review 1. While this is an obvious gap in the literature, the tentative nature of the evidence of effectiveness suggests that further work is needed to establish evidence of effectiveness before cost-effective studies will provide meaningful results. However, costs should be included in intervention trials and there may be scope for modelling of potential cost-savings in terms of cost-effectiveness as well as economic modelling to include broader societal costs such as education, parent employment and incremental savings as CYP grow up.

Due to the apparent importance of social support to the emotional wellbeing of CYP with an LTC highlighted in Review 2, a systematic review that examined the effectiveness and experiences of social support interventions for CYP with LTCs could be valuable. The review could expand on the conceptualisation of social support discussed within this review by considering the model used by Gaysynsky and colleagues.[216] It could potentially consider which methods of social support are experienced as most useful and effective in improving mental health within this population and effective ways of incorporating it into care for CYP with LTCs, and could inform the design of future interventions and studies of their effectiveness.

Involving CYP in an advisory group that met throughout the course of the project to advise on methods, preliminary findings and assist with dissemination material was considered a clear strength of the project and it is recommended that future systematic reviews form an advisory group of true end users wherever possible..

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Closing messages from children and young people advisory group

"Before my diagnosis of SLE at the age of 9 (I am now 17), my life was relatively normal. For the past 9 years I have battled with having to take daily medications; continual check-ups; and various mental health problems mainly around anxiety and depression.

Over the years my biggest struggles have been the lack of control I have felt over my physical and mental health. Both these things affected my ability to socialise; go to school and college consistently; and take part in activities that other teenagers of my age might do. At times this has made life difficult and isolating, with professionals around me not really listening to me.

Being part of this research project has been important, because over the years I have used mental health services with varying affect and they have felt forced rather than a choice. I hope the findings in the research will enable future interventions to be more creative for all young people but particularly for those like me who have long term illnesses. It felt good to think that my experiences and contributions can make a difference."

"Living with a long term medical condition is often much harder than many people think it is. Not only do you have to cope with the health condition itself, but you also have to tackle all the other issues it brings along with it, such as psychological disorders or simply chronic pain, and in a young teenager's normal life of long, tiring days and constantly fluctuating moods, these are enough to really push you over the edge.

I think one of the main issues concerning this are those of awareness and flexibility within conventional activities in a young person's life, such as school. Many teachers in my time at the school have struggled to adjust their policies to my condition. This is the same with many young people coping with long term health conditions

I think researchers need to develop effective methods of improving mental health, and these need to be implemented in schools, as well as outside. This would have a big impact on the young

person's welfare. Perhaps teachers should be better informed about young people's conditions and taught how to support them. Tackling mental health problems before they become too overwhelming, would be beneficial to the young person's physical health, and life in general. Better, more effective methods of intervention need to be found, and be readily accessible. With more research and better awareness in general, the problem of mental health difficulties in children with a long term health condition, can be easily solved."

"I was diagnosed with Juvenile Dermatomyositis in April 2015, which is inflammation of the skin and weakness of the muscles. This results in daily life being very difficult for me because I'm stiff getting out of bed every morning, I struggle climbing stairs, getting out of a bath and walking and standing is very tough as well. I haven't had a complete pain free day in over 2 years.

One of the drugs that I have to take is Methotrexate which is a type of chemotherapy which is meant to reduce the inflammation. I have to inject this into me every Friday night, but every week it makes me feel very nauseas and sick. Every Saturday I feel sick and I can't enjoy my weekend like everybody else and I can miss out on meeting up with family or friends. At first, I was very apprehensive about injecting it into myself because every Friday night I would get myself so worked up and feel very sick, but now I've just learnt to accept it and it has become a part of my routine. However, Methotrexate doesn't help with the pain. So, they say that I have Chronic Pain Syndrome, where I'm always in constant pain.

At the moment, I'm in year 13 and will soon be completing my A Levels, so the stress of the exams, revision and feeling sick from Methotrexate hasn't helped with my pain or my mood. I was diagnosed with depression in January 2017 because for about 6 months prior to this I have been feeling very low and have just felt like I'd had enough because of the stress with school, the pressure of not knowing what I want to do once I'd finished my A Levels and the hospital kept telling me the way to deal with Chronic Pain Syndrome was to have good sleep, to eat, drink, take part in mindfulness and do exercise. I was doing all this but I'm still in so much pain and no one seemed to understand and doctors just say I would feel better in time, but time has no end.

I believe that researchers should work to develop more treatment to help with depression because when you're diagnosed with a long term physical condition, no one tells you that you

could be at risk of developing a mental health problem because you deal with so much; the condition, the medication, the side effects of the medication, the hospital appointments and missing school due to the hospital appointments."

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Contribution of authors

Darren A Moore, Research Fellow, involved in the design and conception of the project, coordinated review work, led reviews and the overarching synthesis, wrote and edited the final report, planned and conducted end user involvement and consultation events.

Michael Nunns, Research Fellow, led Review 1 and the overarching synthesis, wrote and edited the final report, planned and conducted the end user involvement and consultation events.

Liz Shaw, Associate Research Fellow, led Review 2 and the overarching synthesis, wrote and edited the final report, planned and conducted the end user involvement and consultation events.

Morwenna Rogers, Information Specialist, involved in the design and conception of the project, designed and conducted searches for Review 1, Review 2 and economic studies. Retrieval of records for screening, title and abstract screening, edited draft chapters.

Erin Walker, Patient and Public Involvement Lead, coordinated children and young people advisory group, organised young people and parent meetings, conducted end user involvement and consultation events, edited draft chapters.

Tamsin J Ford, Professor, involved in the design and conception of the project, provided mental health topic advice, edited draft chapters.

Ruth Garside, Senior Lecturer, involved in the design and conception of the project, provided qualitative synthesis topic advice, assisted in qualitative synthesis, edited Review 2 draft chapter.

Obioha Ukoumunne, Associate Professor, provided quantitative synthesis topic advice, assisted in quantitative synthesis, edited draft chapters.

Penny Titman, Consultant Clinical Psychologist, involved in the design and conception of the project, provided mental and physical health topic advice, edited draft chapters.

Roz Shafran, Professor, involved in the design and conception of the project, provided mental health topic advice, edited draft chapters.

Isobel Heyman, Professor, involved in the design and conception of the project, provided mental health topic advice, conducted a consultation event, edited draft chapters.

Rob Anderson, Associate Professor, provided health economic topic advice, wrote economic evaluation section, edited draft chapters.

Chris Dickens, Professor, provided mental health topic advice, edited draft chapters.

Russell Viner, Professor, provided paediatric topic advice, edited draft chapters.

Sophie Bennett, Research Associate, involved in the design and conception of the project, provided mental health topic advice, conducted a consultation event, edited overarching synthesis draft chapter.

Stuart Logan, Professor, provided paediatric topic advice, edited overarching synthesis draft chapter.

Fiona Lockhart, Parent advisory group member, involved in the design and conception of the project, attended team meetings and consultation events, wrote foreword and edited plain English summary.

Jo Thompson Coon, Associate Professor, provided overall supervision of the project, involved in the design and conception of the project, provided evidence synthesis topic advice, edited draft chapters, planned and conducted end user involvement and consultation events.

Data sharing statement

This is a systematic review and, therefore, the data used for each analysis are present within the report or supplementary materials.

Further information and requests for access to the data can be obtained from the corresponding author.

References

1. van der Lee JH, Mokkink LB, Grootenhuis MA, Heymans HS, Offringa M. Definitions and measurement of chronic health conditions in childhood: A systematic review. *JAMA* 2007;**297**:2741-51. http://dx.doi.org/10.1001/jama.297.24.2741
2. Perrin EC, Newacheck P, Pless IB, Drotar D, Gortmaker SL, Leventhal J*, et al.* Issues involved in the definition and classification of chronic health conditions. *Pediatrics* 1993;**91**:787-93.
3. Stein RE, Bauman LJ, Westbrook LE, Coupey SM, Ireys HT. Framework for identifying children who have chronic conditions: the case for a new definition. *The Journal of pediatrics* 1993;**122**:342-7.
4. Pless IB, Douglas JW. Chronic illness in childhood: Part I. Epidemiological and clinical characteristics. *Pediatrics* 1971;**47**:405-14.
5. Keyes CL. Mental illness and/or mental health? Investigating axioms of the complete state model of health. *J Consult Clin Psychol* 2005;**73**:539.
6. Nastasi BK, Borja AP. *International handbook of psychological well-being in children and adolescents*. New York, NY: Springer; 2016.
7. Newacheck PW, Taylor WR. Childhood chronic illness: prevalence, severity, and impact. *Am J Public Health* 1992;**82**:364-71. http://dx.doi.org/10.2105/AJPH.88.4.610
8. Brooks F MJ, Klemera E, Chester K, Spencer N, Smeeton N. *HBSC England National Report: Health Behaviour in School-aged Children (HBSC): World Health Organization Collaborative Cross National Study.* Hatfield: University of Hertfordshire; 2015.
9. Denny S, de Silva M, Fleming T, Clark T, Merry S, Ameratunga S*, et al.* The prevalence of chronic health conditions impacting on daily functioning and the association with emotional well-being among a national sample of high school students. *J Adolesc Health* 2014;**54**:410-5. http://dx.doi.org/10.1016/j.jadohealth.2013.09.010
10. Boyce WF, Davies D, Raman SR, Tynjala J, Valimaa R, King M*, et al.* Emotional health of Canadian and Finnish students with disabilities or chronic conditions. *Int J Rehabil Res* 2009;**32**:154-61. http://dx.doi.org/10.1097/MRR.0b013e32831e452e
11. Newacheck PW, Halfon N. Prevalence and impact of disabling chronic conditions in childhood. *Am J Public Health* 1998;**88**:610-7.
12. Dey M, Wang J, Jorm AF, Mohler-Kuo M. Children with mental versus physical health problems: differences in perceived disease severity, health care service utilization and parental health literacy. *Soc Psychiatry Psychiatr Epidemiol* 2015;**50**:407-18. http://dx.doi.org/10.1007/s00127-014-0944-7
13. Hysing M, Elgen I, Gillberg C, Lie SA, Lundervold AJ. Chronic physical illness and mental health in children. Results from a large-scale population study. *J Child Psychol Psychiatry* 2007;**48**:785-92. http://dx.doi.org/10.1111/j.1469-7610.2007.01755.x
14. Pinquart M, Shen Y. Depressive symptoms in children and adolescents with chronic physical illness: an updated meta-analysis. *J Pediatr Psychol* 2011;**36**:375-84. http://dx.doi.org/10.1093/jpepsy/jsq104
15. Gupta V, Singh A, Singh TB, Upadhyay S. Psychological morbidity in children undergoing chemotherapy for acute lymphoblastic leukemia. *Indian J Pediatr* 2014;**81**:699-701. http://dx.doi.org/10.1007/s12098-013-1211-8

16. Esmaeeli MR, Erfani Sayar R, Saghebi A, Elmi S, Rahmani S, Elmi S*, et al.* Screening for depression in hospitalized pediatric patients. *Iran J Child Neurol* 2014;**8**:47-51.
17. Kogon AJ, Vander Stoep A, Weiss NS, Smith J, Flynn JT, McCauley E. Depression and its associated factors in pediatric chronic kidney disease. *Pediatr Nephrol* 2013;**28**:1855-61. http://dx.doi.org/10.1007/s00467-013-2497-5
18. Merikangas KR, Calkins ME, Burstein M, He JP, Chiavacci R, Lateef T*, et al.* Comorbidity of physical and mental disorders in the neurodevelopmental genomics cohort study. *Pediatrics* 2015;**135**:e927-38. http://dx.doi.org/10.1542/peds.2014-1444
19. Katon W, Lozano P, Russo J, McCauley E, Richardson L, Bush T. The prevalence of DSM-IV anxiety and depressive disorders in youth with asthma compared with controls. *J Adolesc Health* 2007;**41**:455-63.
20. Acosta-Perez E, Canino G, Ramirez R, Prelip M, Martin M, Ortega AN. Do Puerto Rican youth with asthma and obesity have higher odds for mental health disorders? *Psychosomatics* 2012;**53**:162-71. http://dx.doi.org/10.1016/j.psym.2011.07.011
21. Greenley RN, Hommel KA, Nebel J, Raboin T, Li SH, Simpson P*, et al.* A meta-analytic review of the psychosocial adjustment of youth with inflammatory bowel disease. *J Pediatr Psychol* 2010;**35**:857-69. http://dx.doi.org/10.1093/jpepsy/jsp120
22. Shelby GD, Shirkey KC, Sherman AL, Beck JE, Haman K, Shears AR*, et al.* Functional abdominal pain in childhood and long-term vulnerability to anxiety disorders. *Pediatrics* 2013;**132**:475-82. http://dx.doi.org/10.1542/peds.2012-2191
23. Moreira JM, Bouissou Morais Soares CM, Teixeira AL, Simoes ESAC, Kummer AM. Anxiety, depression, resilience and quality of life in children and adolescents with pre-dialysis chronic kidney disease. *Pediatr Nephrol* 2015;**30**:2153-62. http://dx.doi.org/10.1007/s00467-015-3159-6
24. Kogon AJ, Matheson MB, Flynn JT, Gerson AC, Warady BA, Furth SL*, et al.* Depressive symptoms in children with chronic kidney disease. *J Pediatr* 2016;**168**:164-70. http://dx.doi.org/10.1016/j.jpeds.2015.09.040
25. Rousseau-Salvador C, Amouroux R, Annequin D, Salvador A, Tourniaire B, Rusinek S. Anxiety, depression and school absenteeism in youth with chronic or episodic headache. *Pain Res Manag* 2014;**19**:235-40. http://dx.doi.org/10.1155/2014/541618
26. Wändell P, Ljunggren G, Wahlström L, Carlsson AC. Diabetes and psychiatric illness in the total population of Stockholm. *J Psychosom Res* 2014;**77**:169-73. http://dx.doi.org/10.1016/j.jpsychores.2014.06.012
27. Plioplys S, Dunn DW, Caplan R. 10-year research update review: psychiatric problems in children with epilepsy. *J Am Acad Child Adolesc Psychiatry* 2007;**46**:1389-402. http://dx.doi.org/10.1097/chi.0b013e31815597fc
28. Wechsler A, Sánchez-Iglesias I. Psychological adjustment of children with cancer as compared with healthy children: a meta-analysis. *European journal of cancer care* 2013;**22**:314-25. http://dx.doi.org/10.1111/ecc.12031
29. Arabiat DH, Elliott B, Draper P. The prevalence of depression in pediatric oncology patients undergoing chemotherapy treatment in Jordan. *J Pediatr Oncol Nurs* 2012;**29**:283-8. http://dx.doi.org/10.1177/1043454212451524
30. Myers RM, Balsamo L, Lu X, Devidas M, Hunger SP, Carroll WL*, et al.* A prospective study of anxiety, depression, and behavioral changes in the first year after a diagnosis of childhood

acute lymphoblastic leukemia: a report from the Children's Oncology Group. *Cancer* 2014;**120**:1417-25. http://dx.doi.org/10.1002/cncr.28578
31. von Essen L, Enskar K, Kreuger A, Larsson B, Sjoden PO. Self-esteem, depression and anxiety among Swedish children and adolescents on and off cancer treatment. *Acta Paediatr* 2000;**89**:229-36. http://dx.doi.org/10.1111/j.1651-2227.2000.tb01221.x
32. Chang G, Ratichek SJ, Recklitis C, Syrjala K, Patel SK, Harris L*, et al.* Children's psychological distress during pediatric HSCT: parent and child perspectives. *Pediatr Blood Cancer* 2012;**58**:289-96. http://dx.doi.org/10.1002/pbc.23185
33. Phipps S, Dunavant M, Lensing S, Rai S. Acute health-related quality of life in children undergoing stem cell transplant: II. Medical and demographic determinants. *Bone Marrow Transplant* 2002;**29**:435. http://dx.doi.org/.10.1038/sj.bmt.1703377
34. Beyenburg S, Mitchell AJ, Schmidt D, Elger CE, Reuber M. Anxiety in patients with epilepsy: systematic review and suggestions for clinical management. *Epilepsy Behav* 2005;**7**:161-71. http://dx.doi.org/10.1016/j.yebeh.2005.05.014
35. Golden SH, Lazo M, Carnethon M, Bertoni AG, Schreiner PJ, Roux AVD*, et al.* Examining a bidirectional association between depressive symptoms and diabetes. *JAMA* 2008;**299**:2751-9. http://dx.doi.org/10.1001/jama.299.23.2751
36. Wallander JL, Varni JW, Babani L, Banis HT, Wilcox KT. Family resources as resistance factors for psychological maladjustment in chronically ill and handicapped children. *J Pediatr Psychol* 1989;**14**:157-73.
37. Wallander JL, Varni JW. Adjustment in children with chronic physical disorders: programmatic research on a disability-stress-coping model. In: La Greca AM, Siegel LJ, Wallander JL, Walker CE, editors. *Stress and coping in child health*New York, NY: Guilford Press; 1992:279-98.
38. Thompson R, Gustafson K. Psychological adjustment of children with chronic illness. *Adaptation to chronic childhood illness Washington, DC: American Psychological Association* 1996:57-86.
39. Thompson RJ, Gustafson KE, Gil KM, Kinney TR, Spock A. Change in the psychological adjustment of children with cystic fibrosis or sickle cell disease and their mothers. *J Clin Psychol Med Settings* 1999;**6**:373-92. https://dx.doi.org/10.1023/A:1026219831205
40. Moos RH, Holahan CJ. Adaptive tasks and methods of coping with illness and disability. In: Martz E, Livneh H, editors. *Coping with chronic illness and disability*New York, NY: Springer; 2007:107-26.
41. Haase JE, Kintner EK, Monahan PO, Robb SL. The resilience in illness model, part 1: exploratory evaluation in adolescents and young adults with cancer. *Cancer Nurs* 2014;**37**:E1-12. http://dx.doi.org/10.1097/NCC.0b013e31828941bb
42. Gannoni AF, Shute RH. Parental and child perspectives on adaptation to childhood chronic illness: a qualitative study. *Clin Child Psychol Psychiatry* 2010;**15**:39-53. http://dx.doi.org/10.1177/1359104509338432
43. Christie D, Romano G, Barnes J, Madge N, Nicholas DB, Koot HM*, et al.* Exploring views on satisfaction with life in young children with chronic illness: an innovative approach to the collection of self-report data from children under 11. *Clin Child Psychol Psychiatry* 2012;**17**:5-15. http://dx.doi.org/10.1177/1359104510392309

44. Venning A, Eliott J, Wilson A, Kettler L. Understanding young peoples' experience of chronic illness: a systematic review. *International Journal of Evidence-Based Healthcare* 2008;**6**:321-36. https://dx.doi.org/10.1111/j.1744-1609.2008.00107.x
45. Cottrell D. Prevention and treatment of psychiatric disorders in children with chronic physical illness. *Arch Dis Child* 2015;**100**:303-4. http://dx.doi.org/10.1136/archdischild-2014-307866
46. Lustman PJ, Clouse RE. Depression in diabetic patients: the relationship between mood and glycemic control. *J Diabetes Complications* 2005;**19**:113-22. http://dx.doi.org/10.1016/j.jdiacomp.2004.01.002
47. Chavira DA, Garland AF, Daley S, Hough R. The impact of medical comorbidity on mental health and functional health outcomes among children with anxiety disorders. *Journal of developmental and behavioral pediatrics: JDBP* 2008;**29**:394.
48. Kovacs M, Mukerji P, Drash A, Iyengar S. Biomedical and psychiatric risk factors for retinopathy among children with IDDM. *Diabetes Care* 1995;**18**:1592-9.
49. Naylor C, Parsonage M, McDaid D, Knapp M, Fossey M, Galea A. *Long-term conditions and mental health: the cost of co-morbidities*: The King's Fund; 2012.
50. Chiles JA, Lambert MJ, Hatch AL. The impact of psychological interventions on medical cost offset: A meta-analytic review. *Clinical Psychology: Science and Practice* 1999;**6**:204-20.
51. Chorpita BF, Daleiden EL, Ebesutani C, Young J, Becker KD, Nakamura BJ*, et al.* Evidence-based treatments for children and adolescents: An updated review of indicators of efficacy and effectiveness. *Clinical Psychology: Science and Practice* 2011;**18**:154-72. http://dx.doi.org/10.1111/j.1468-2850.2011.01247.x
52. Excellence NIfHaC. *Antisocial behaviour and conduct disorders in children and young people: recognition and management*. London: NICE; 2013.
53. Excellence NIfHaC. *Depression in children and young people: identification and management* London: NICE; 2005.
54. Kendall PC, Hudson JL, Gosch E, Flannery-Schroeder E, Suveg C. Cognitive-behavioral therapy for anxiety disordered youth: a randomized clinical trial evaluating child and family modalities. *J Consult Clin Psychol* 2008;**76**:282. http://dx.doi.org/http://psycnet.apa.org/doi/10.1037/0022-006X.76.2.282
55. Johnson B, Eiser C, Young V, Brierley S, Heller S. Prevalence of depression among young people with Type 1 diabetes: a systematic review. *Diabet Med* 2013;**30**:199-208. http://dx.doi.org/10.1111/j.1464-5491.2012.03721.x
56. Markowitz SM, Gonzalez JS, Wilkinson JL, Safren SA. A review of treating depression in diabetes: emerging findings. *Psychosomatics* 2011;**52**:1-18. http://dx.doi.org/10.1016/j.psym.2010.11.007
57. Ekinci O, Titus JB, Rodopman AA, Berkem M, Trevathan E. Depression and anxiety in children and adolescents with epilepsy: prevalence, risk factors, and treatment. *Epilepsy Behav* 2009;**14**:8-18. http://dx.doi.org/10.1016/j.yebeh.2008.08.015
58. Szigethy EVA, Whitton SW, Levy-Warren A, DeMaso DR, Weisz J, Beardslee WR. Cognitive-Behavioral Therapy for Depression in Adolescents With Inflammatory Bowel Disease: A Pilot Study. *J Am Acad Child Adolesc Psychiatry* 2004;**43**:1469-77. http://dx.doi.org/10.1097/01.chi.0000142284.10574.1f

59. Richardson LP, Russo JE, Lozano P, McCauley E, Katon W. The effect of comorbid anxiety and depressive disorders on health care utilization and costs among adolescents with asthma. *Gen Hosp Psychiatry* 2008;**30**:398-406.
60. Health Do. *No Health Without Mental Health: A cross-government outcomes strategy*. London: Department of Health; 2011.
61. Confederation N. *Investing in emotional and psychological wellbeing for patients with long-term conditions*. London: The NHS Confederation; 2012.
62. Excellence. NIfHaC. *Diabetes (type 1 and type 2) in children and young people: diagnosis and management*. London: NICE; 2015.
63. Excellence. NIfHaC. *Epilepsies: Diagnosis and management*. London: NICE; 2012.
64. Social Care LGaCPD. *Closing the Gap: Priorities for essential change in mental health*. London: Department of Health; 2014.
65. Taskforce CaYPsMH. *Future in mind: Promoting, protecting and improving our children and young people's mental health and wellbeing*. London: Department of Health; 2015.
66. Excellence. NIfHaC. *Depression in adults with a chronic physical health problem: recognition and management*. London: NICE; 2009.
67. Sharpe M, Walker J, Hansen CH, Martin P, Symeonides S, Gourley C*, et al.* Integrated collaborative care for comorbid major depression in patients with cancer (SMaRT Oncology-2): a multicentre randomised controlled effectiveness trial. *The Lancet* 2014;**384**:1099-108.
68. Owens PL, Hoagwood K, Horwitz SM, Leaf PJ, Poduska JM, Kellam SG*, et al.* Barriers to children's mental health services. *J Am Acad Child Adolesc Psychiatry* 2002;**41**:731-8.
69. James AC, James G, Cowdrey FA, Soler A, Choke A. Cognitive behavioural therapy for anxiety disorders in children and adolescents. *Cochrane Database of Systematic Reviews* 2015;**6**. http://dx.doi.org/10.1002/14651858.CD004690.pub3
70. Gertler P, Tate RL, Cameron ID. Non-pharmacological interventions for depression in adults and children with traumatic brain injury. *Cochrane Database of Systematic Reviews* 2015;**6**. http://dx.doi.org/10.1002/14651858.CD004690.pub3
71. Lane DA, Millane TA, Lip GY. Psychological interventions for depression in adolescent and adult congenital heart disease. *Cochrane database of systematic reviews* 2013;**10**. http://dx.doi.org/10.1002/14651858.CD004372.pub2
72. Eccleston C, Palermo TM, Fisher E, Law E. Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database of Systematic Reviews* 2012;**8**. http://dx.doi.org/10.1002/14651858.CD009660.pub2
73. Glasscoe CA, Quittner AL. Psychological interventions for people with cystic fibrosis and their families. *Cochrane Database of Systematic Reviews* 2008;**3**. http://dx.doi.org/10.1002/14651858.CD003148.pub2
74. Ramaratnam S, Baker GA, Goldstein LH. Psychological treatments for epilepsy. *Cochrane Database of Systematic Reviews* 2008;**3**. http://dx.doi.org/10.1002/14651858.CD002029.pub3
75. Timmer A, Preiss JC, Motschall E, Rücker G, Jantschek G, Moser G. Psychological interventions for treatment of inflammatory bowel disease. *Cochrane Database of Systematic Reviews* 2011;**2**. http://dx.doi.org/10.1002/14651858.CD006913.pub2
76. Gandy M, Sharpe L, Perry KN. Cognitive behavior therapy for depression in people with epilepsy: a systematic review. *Epilepsia* 2013;**54**:1725-34.
77. Yorke J, Fleming SL, Shuldham C. A systematic review of psychological interventions for children with asthma. *Pediatr Pulmonol* 2007;**42**:114-24.

78. Sansom-Daly UM, Peate M, Wakefield CE, Bryant RA, Cohn RJ. A systematic review of psychological interventions for adolescents and young adults living with chronic illness. *Health Psychol* 2012;**31**:380-93. http://dx.doi.org/10.1037/a0025977
79. Thompson RD, Delaney P, Flores I, Szigethy E. Cognitive-behavioral therapy for children with comorbid physical illness. *Child Adolesc Psychiatr Clin N Am* 2011;**20**:329-48.
80. Thomé-Souza M, Kuczynski E, Valente K. Sertraline and fluoxetine: safe treatments for children and adolescents with epilepsy and depression. *Epilepsy Behav* 2007;**10**:417-25.
81. Bennett S, Shafran R, Coughtrey A, Walker S, Heyman I. Psychological interventions for mental health disorders in children with chronic physical illness: a systematic review. *Arch Dis Child* 2015;**100**:308-16. http://dx.doi.org/10.1136/archdischild-2014-307474
82. Martinović Ž, Simonović P, Djokić R. Preventing depression in adolescents with epilepsy. *Epilepsy Behav* 2006;**9**:619-24.
83. Szigethy E, Kenney E, Carpenter J, Hardy DM, Fairclough D, Bousvaros A*, et al.* Cognitive-behavioral therapy for adolescents with inflammatory bowel disease and subsyndromal depression. *J Am Acad Child Adolesc Psychiatry* 2007;**46**:1290-8.
84. dissemination Cfra. *Systematic reviews: CRD's guidance for undertaking reviews in health care*: Centre for Reviews and Dissemination; 2009.
85. Higgins JP, Green S. *Cochrane handbook for systematic reviews of interventions*: John Wiley & Sons; 2011.
86. Glanville JM, Lefebvre C, Miles JN, Camosso-Stefinovic J. How to identify randomized controlled trials in MEDLINE: ten years on. *J Med Libr Assoc* 2006;**94**:130.
87. Eccleston C, Palermo TM, Williams AC, Lewandowski Holley A, Morley S, Fisher E*, et al.* Psychological therapies for the management of chronic and recurrent pain in children and adolescents. *Cochrane Database of Systematic Reviews* 2014;**5**. http://dx.doi.org/10.1002/14651858.CD003968.pub4
88. Canter KS, Roberts MC. A systematic and quantitative review of interventions to facilitate school reentry for children with chronic health conditions. *J Pediatr Psychol* 2012;**37**:1065-75.
89. Taylor SJ, Pinnock H, Epiphaniou E, Pearce G, Parke HL, Schwappach A*, et al.* A rapid synthesis of the evidence on interventions supporting self-management for people with long-term conditions: PRISMS–Practical systematic Review of Self-Management Support for long-term conditions. *Health Services and Delivery Research* 2014;**2**. http://dx.doi.org/10.3310/hsdr02530
90. Sattoe JN, Bal MI, Roelofs PD, Bal R, Miedema HS, van Staa A. Self-management interventions for young people with chronic conditions: A systematic overview. *Patient Educ Couns* 2015;**98**:704-15. http://dx.doi.org/10.1016/j.pec.2015.03.004
91. Fisher E, Law E, Palermo TM, Eccleston C. Psychological therapies (remotely delivered) for the management of chronic and recurrent pain in children and adolescents. *Cochrane Database Syst Rev* 2015;**3**:Cd011118. http://dx.doi.org/10.1002/14651858.CD011118.pub2
92. Galdas P, Fell J, Bower P, Kidd L, Blickem C, McPherson K*, et al.* The effectiveness of self-management support interventions for men with long-term conditions: a systematic review and meta-analysis. *BMJ open* 2015;**5**:e006620.
93. Lambert V, Keogh D. Striving to live a normal life: a review of children and young people's experience of feeling different when living with a long term condition. *J Pediatr Nurs* 2015;**30**:63-77. http://dx.doi.org/10.1016/j.pedn.2014.10.012

10.1016/j.pedn.2014.09.016

94. Schwartz L, Drotar D. Defining the nature and impact of goals in children and adolescents with a chronic health condition: A review of research and a theoretical framework. *J Clin Psychol Med Settings* 2006;**13**:390-402.
95. Spencer NJ, Blackburn CM. Disabling chronic conditions in childhood and socioeconomic disadvantage: a systematic review and meta-analyses of observational studies. 2015;**5**:e007062. http://dx.doi.org/10.1136/bmjopen-2014-007062
96. Braam KI, van der Torre P, Takken T, Veening MA, van Dulmen-den Broeder E, Kaspers GJ. Physical exercise training interventions for children and young adults during and after treatment for childhood cancer. *Cochrane Database Syst Rev* 2013;**4**:Cd008796. http://dx.doi.org/10.1002/14651858.CD008796.pub2
97. Nightingale R, Friedl S, Swallow V. Parents' learning needs and preferences when sharing management of their child's long-term/chronic condition: A systematic review. *Patient Educ Couns* 2015;**98**:1329-38.
98. Smith J, Cheater F, Bekker H. Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature. *Health Expect* 2015;**18**:452-74. http://dx.doi.org/10.1111/hex.12040
99. Orrskog S, Medin E, Tsolova S, Semenza JC. Causal inference regarding infectious aetiology of chronic conditions: a systematic review. *PLoS One* 2013;**8**:e68861.
100. Ludvigsen MS, Fegran L, Aagaard H, Meyer G, Uhrenfeldt L. Parents' experiences of the transfer from pediatric to adult care for their adolescent or young adult children with chronic conditions: a systematic review protocol. *JBI Database of Systematic Reviews and Implementation Reports* 2014;**12**:102-11.
101. Quiñones A, Richardson J, Freeman M, O'Neil ME, Kansagara D. Group Visits Focusing on Education for the Management of Chronic Conditions in Adults: A Systematic Review. *VA-ESP Project #05-225* 2012.
102. Brown CA, Kuo M, Phillips L, Berry R, Tan M. Non-pharmacological sleep interventions for youth with chronic health conditions: a critical review of the methodological quality of the evidence. *Disabil Rehabil* 2013;**35**:1221-55. http://dx.doi.org/10.3109/09638288.2012.723788
103. Hardelid P, Dattani N, Gilbert R. Estimating the prevalence of chronic conditions in children who die in England, Scotland and Wales: a data linkage cohort study. *BMJ open* 2014;**4**:e005331.
104. Feudtner C, Feinstein JA, Zhong W, Hall M, Dai D. Pediatric complex chronic conditions classification system version 2: updated for ICD-10 and complex medical technology dependence and transplantation. *BMC Pediatr* 2014;**14**:199.
105. Glanville J, Kaunelis D, Mensinkai S. How well do search filters perform in identifying economic evaluations in MEDLINE and EMBASE. *Int J Technol Assess Health Care* 2009;**25**:522-9.
106. Richardson M, Moore D, Gwernan-Jones R, Thompson-Coon J, Ukoumunne O, Rogers M*, et al.* Non-pharmacological interventions for attention-deficit/hyperactivity disorder (ADHD) delivered in school settings: systematic reviews of quantitative and qualitative research. *Health technology assessment (Winchester, England)* 2015;**19**:1-470.
107. Cohen J. A power primer. *Psychol Bull* 1992;**112**:155.
108. Glass GV, McGaw B, Smith M. *Meta-analysis in social research.* . Beverly Hills, CA:: Sage; 1981.

109. Higgins JP, Thompson SG, Deeks JJ, Altman DG. Measuring inconsistency in meta-analyses. *BMJ* 2003;**327**:557-60.
110. Borenstein M, Hedges LV, Higgins JP, Rothstein HR. *Introduction to Meta-Analysis*. Chichester, UK: Wiley; 2009.
111. Ahmed I, Sutton AJ, Riley RD. Assessment of publication bias, selection bias, and unavailable data in meta-analyses using individual participant data: a database survey. *BMJ* 2012;**344**:d7762.
112. Moher D, Liberati A, Tetzlaff J, Altman DG, Group P. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med* 2009;**6**:e1000097.
113. Whittemore R, Grey M, Lindemann E, Ambrosino J, Jaser S. Development of an Internet coping skills training program for teenagers with type 1 diabetes. *CIN: Computers, Informatics, Nursing* 2010;**28**:103-11. http://dx.doi.org/10.1097/NCN.0b013e3181cd8199
114. Foundation H. *Doing diabetes differently: a noval way of contracting integrated psychological therapies to increase treatment adherence*. Milton Keynes: Health Foundation; 2014.
115. Foster EM, Prinz RJ, Sanders MR, Shapiro CJ. The costs of a public health infrastructure for delivering parenting and family support. *Children and Youth Services Review* 2008;**30**:493-501.
116. Mihalopoulos C, Sanders MR, Turner KM, Murphy-Brennan M, Carter R. Does the Triple P–Positive Parenting Program provide value for money? *Aust N Z J Psychiatry* 2007;**41**:239-46.
117. Westrupp EM, Northam E, Lee KJ, Scratch S, Cameron F. Reducing and preventing internalizing and externalizing behavior problems in children with type 1 diabetes: A randomized controlled trial of the Triple P-Positive Parenting Program. *Pediatr Diabetes* 2015;**16**:554-63. http://dx.doi.org/10.1111/pedi.12205
118. Bignall WJR, Luberto CM, Cornette AF, Haj-Hamed M, Cotton S. Breathing retraining for African-American adolescents with asthma: A pilot study of a school-based randomized controlled trial. *J Asthma* 2015;**52**:889-96. http://dx.doi.org/10.3109/02770903.2015.1033724
119. Boardway RH, Delamater AM, Tomakowsky J, Gutai JP. Stress management training for adolescents with diabetes. *J Pediatr Psychol* 1993;**18**:29-45.
120. Diego MA, Field T, Hernandez-Reif M, Shaw K, Friedman L, Ironson G. HIV adolescents show improved immune function following massage therapy. *Int J Neurosci* 2001;**106**:35-45.
121. Hains AA, Davies WH, Parton E, Totka J, Amoroso-Camarata J. A stress management intervention for adolescents with type 1 diabetes. *The Diabetes educator* 2000;**26**:417-24.
122. Lyon ME, Jacobs S, Briggs L, Cheng YI, Wang J. A longitudinal, randomized, controlled trial of advance care planning for teens with cancer: Anxiety, depression, quality of life, advance directives, spirituality. *J Adolesc Health* 2014;**54**:710-7. http://dx.doi.org/10.1016/j.jadohealth.2013.10.206
123. Masia Warner C, Colognori D, Kim RE, Reigada LC, Klein RG, Browner-Elhanan KJ*, et al.* Cognitive-behavioral treatment of persistent functional somatic complaints and pediatric anxiety: An initial controlled trial. *Depress Anxiety* 2011;**28**:551-9. http://dx.doi.org/10.1002/da.20821
124. Reigada LC, Polokowski AR, Walder DJ, Szigethy EM, Benkov KJ, Bruzzese J-M*, et al.* Treatment for comorbid pediatric gastrointestinal and anxiety disorders: A pilot study of a flexible health sensitive cognitive-behavioral therapy program. *Clinical Practice in Pediatric Psychology* 2015;**3**:314-26. http://dx.doi.org/10.1037/cpp0000116

125. Szigethy E, Bujoreanu SI, Youk AO, Weisz J, Benhayon D, Fairclough D*, et al.* Randomized efficacy trial of two psychotherapies for depression in youth with inflammatory bowel disease. *J Am Acad Child Adolesc Psychiatry* 2014;**53**:726-35. http://dx.doi.org/10.1016/j.jaac.2014.04.014
126. Yetwin AK. *Heart Rate Variability Biofeedback Therapy for Children and Adolescents with Chronic Pain*: Alliant International University; 2011.
127. Brown FL, Whittingham K, Boyd RN, McKinlay L, Sofronoff K. Improving child and parenting outcomes following paediatric acquired brain injury: a randomised controlled trial of Stepping Stones Triple P plus Acceptance and Commitment Therapy. *Journal of child psychology and psychiatry* 2014;**55**:1172-83.
128. Gordon BA, Knapman LM, Lubitz L. Graduated exercise training and progressive resistance training in adolescents with chronic fatigue syndrome: a randomized controlled pilot study. *Clin Rehabil* 2010;**24**:1072-9.
129. Serlachius AS, Scratch SE, Northam EA, Frydenberg E, Lee KJ, Cameron FJ. A randomized controlled trial of cognitive behaviour therapy to improve glycaemic control and psychosocial wellbeing in adolescents with type 1 diabetes. *J Health Psychol* 2014:1359105314547940.
130. Whittingham K, Sanders M, McKinlay L, Boyd RN. Interventions to reduce behavioral problems in children with cerebral palsy: an RCT. *Pediatrics* 2014;**133**:e1249-e57. http://dx.doi.org/10.1542/peds.2013-3620
131. Ashori M, Hasanzadeh S, Pourmohamadrezatajrishi M. Effectiveness of life skill instruction on the mental health of hearing loss students. *J Rehabil* 2013;**13**:48-57.
132. Nekah A, Mohsen S, Kamali F, Jansouz F. The Effects of Structured Cognitive-Behavioral Group Play Therapy on Anxiety and Depression in Children with Cancer: A Pilot Study. *Evidence Based Care* 2015;**5**:39-50.
133. Pourmohamadreza-Tajrishi M, Ashori M, Jalilabkenar SS. The Effectiveness of Emotional Intelligence Training on the Mental Health of Male Deaf Students. *Iranian Journal of Public Health* 2013;**42**:1174-80.
134. Zareapour A, Khoshknab MF, Kashaninia A, Biglarian A, Babashahabi R. Effect of group play therapy on depression in children with cancer. *Scientific Journal of Kurdistan University of Medical Sciences* 2009;**14**:64-72.
135. Bufalini A. Role of interactive music in oncological pediatric patients undergoing painful procedures. [Italian]. *Minerva Pediatr* 2009;**61**:379-89.
136. Shoshani A, Mifano K, Czamanski-Cohen J. The effects of the Make a Wish intervention on psychiatric symptoms and health-related quality of life of children with cancer: a randomised controlled trial. *Qual Life Res* 2015;**25**:1209-18. http://dx.doi.org/10.1007/s11136-015-1148-7
137. Wang Q, Zhang J, Mei Q, Huang Y, Liu Y, Yuan X*, et al.* Group play therapy for improving mental coping ability in children with asthma. [Chinese]. *Medical Journal of Chinese People's Liberation Army* 2012;**37**:623-9.
138. Yang HB, Tang YQ, Yi ZW. Effect of relaxation training on psychosomatic symptoms of children with asthma. *Chinese Journal of Clinical Rehabilitation* 2004;**8**:8392-3.
139. Brown FL, Whittingham K, Boyd RN, McKinlay L, Sofronoff K. Does Stepping Stones Triple P plus Acceptance and Commitment Therapy improve parent, couple, and family adjustment following paediatric acquired brain injury? A randomised controlled trial. *Behav Res Ther* 2015;**73**:58-66.

140. Lyon ME, Jacobs S, Briggs L, Cheng Yao I, Wang J. A longitudinal, randomized, controlled trial of advance care planning for teens with cancer: Anxiety, depression, quality of life, advance directives, spirituality. *J Adolesc Health* 2013;**54**:710-7. http://dx.doi.org/10.1016/j.jadohealth.2013.10.206
141. Szigethy E, Craig AE, Iobst EA, Grand RJ, Keljo D, DeMaso D*, et al.* Profile of depression in adolescents with inflammatory bowel disease: Implications for treatment. *Inflamm Bowel Dis* 2009;**15**:69-74. http://dx.doi.org/10.1002/ibd.20693
142. Thompson RD, Craig A, Crawford EA, Fairclough D, Gonzalez-Heydrich J, Bousvaros A*, et al.* Longitudinal results of cognitive behavioral treatment for youths with inflammatory bowel disease and depressive symptoms. *J Clin Psychol Med Settings* 2012;**19**:329-37. http://dx.doi.org/10.1007/s10880-012-9301-8
143. Szigethy E, Youk AO, Gonzalez-Heydrich J, Bujoreanu SI, Weisz J, Fairclough D*, et al.* Effect of 2 psychotherapies on depression and disease activity in pediatric Crohn's disease. *Inflamm Bowel Dis* 2015;**21**:1321-8. http://dx.doi.org/10.1097/MIB.0000000000000358
144. Whittingham K, Sanders MR, McKinlay L, Boyd RN. Parenting Intervention Combined With Acceptance and Commitment Therapy: A Trial With Families of Children With Cerebral Palsy. *J Pediatr Psychol* 2015; 10.1093/jpepsy/jsv118. http://dx.doi.org/10.1093/jpepsy/jsv118
145. Wicksell RK, Melin L, Lekander M, Olsson GL. Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain–a randomized controlled trial. *Pain* 2009;**141**:248-57.
146. Karlson CW, Rapoff MA. Attrition in randomized controlled trials for pediatric chronic conditions. *J Pediatr Psychol* 2008;**34**:782-93.
147. Sanders MR, Mazzucchelli TG, Studman LJ. Stepping Stones Triple P: the theoretical basis and development of an evidence-based positive parenting program for families with a child who has a disability. *Journal of Intellectual and Developmental Disability* 2004;**29**:265-83.
148. Abrahamse ME, Junger M, Leijten PH, Lindeboom R, Boer F, Lindauer RJ. Psychometric properties of the Dutch Eyberg Child Behavior Inventory (ECBI) in a community sample and a multi-ethnic clinical sample. *Journal of psychopathology and behavioral assessment* 2015;**37**:679-91.
149. Ewing B. Wish fulfillment: Palliative care and end-of-life intervention. *Pediatr Nurs* 2009;**35**:81-5.
150. Excellence NIfHC. *Depression in children and young people: Identification and management*: NICE clinical guideline 28; 2005.
151. Kovacs M. CDI. *Multi-Health Systems, Inc, North Tonawanda, NY* 1992; https://doi.org/10.2337/diacare.18.12.1592.
http://dx.doi.org/https://doi.org/10.2337/diacare.18.12.1592
152. Julian LJ. Measures of anxiety: State-Trait Anxiety Inventory (STAI), Beck Anxiety Inventory (BAI), and Hospital Anxiety and Depression Scale-Anxiety (HADS-A). *Arthritis Care Res* 2011;**63**:S467-S72.
153. Challen AR, Machin SJ, Gillham JE. The UK Resilience Programme: A school-based universal nonrandomized pragmatic controlled trial. *J Consult Clin Psychol* 2014;**82**:75.
154. Robb SL, Clair AA, Watanabe M, Monahan PO, Azzouz F, Stouffer JW*, et al.* Randomized controlled trial of the active music engagement (AME) intervention on children with cancer. *Psycho-Oncology* 2008;**17**:699-708.

155. Beebe A, Gelfand EW, Bender B. A randomized trial to test the effectiveness of art therapy for children with asthma. *J Allergy Clin Immunol* 2010;**126**:263-6. e1.
156. Murphy H, Wadham C, Hassler-Hurst J, Rayman G, Skinner T. Randomized trial of a diabetes self-management education and family teamwork intervention in adolescents with Type 1 diabetes. *Diabet Med* 2012;**29**:e249-e54.
157. Subirana M, Solá I, Garcia JM, Gich I, Urrútia G. A nursing qualitative systematic review required MEDLINE and CINAHL for study identification. *J Clin Epidemiol* 2005;**58**:20-5.
158. Wright K, Golder S, Lewis-Light K. What value is the CINAHL database when searching for systematic reviews of qualitative studies? *Systematic reviews* 2015;**4**:104.
159. Wallace A, Croucher K, Quilgars D, Baldwin S. Meeting the challenge: developing systematic reviewing in social policy. *Policy & politics* 2004;**32**:455-70.
160. Moore DA, Gwernan-Jones R, Richardson M, Racey D, Rogers M, Stein K*, et al.* The experiences of and attitudes toward non-pharmacological interventions for attention-deficit/hyperactivity disorder used in school settings: a systematic review and synthesis of qualitative research. *Emotional and Behavioural Difficulties* 2016;**21**:61-82.
161. Noblit GW, Hare RD. *Meta-ethnography: Synthesizing qualitative studies*. Newbury Park, CA: Sage; 1988.
162. Morton K, Dennison L, May C, Murray E, Little P, McManus RJ*, et al.* Using digital interventions for self-management of chronic physical health conditions: A meta-ethnography review of published studies. *Patient Educ Couns* 2016.
163. Evans R, Hurrell C. The role of schools in children and young people's self-harm and suicide: systematic review and meta-ethnography of qualitative research. *BMC Public Health* 2016;**16**:401.
164. Frost J, Garside R, Cooper C, Britten N. A qualitative synthesis of diabetes self-management strategies for long term medical outcomes and quality of life in the UK. *BMC Health Serv Res* 2014;**14**:348.
165. Ames HM, Glenton C, Lewin S. Parents' and informal caregivers' views and experiences of communication about routine childhood vaccination: a synthesis of qualitative evidence. *The Cochrane Library* 2017.
166. Suri H. Purposeful sampling in qualitative research synthesis. *Qualitative Research Journal* 2011;**11**:63-75.
167. Barnetz Z, Feigin R. "We didn't have to talk": Adolescent perception of mentor-mentee relationships in an evaluation study of a mentoring program for adolescents with juvenile diabetes. *Child & Adolescent Social Work Journal* 2012;**29**:463-83. http://dx.doi.org/10.1007/s10560-012-0273-1
168. Bluebond-Langer M, Perkel D, Goertzel T. Pediatric cancer patients' peer relationships: The impact of an oncology camp experience. *J Psychosoc Oncol* 1991;**9**:67-80.
169. Brodeur SS. Treating families coping with chronic illness: An evaluation of the "Living Well" program. *Dissertation Abstracts International: Section B: The Sciences and Engineering* 2005;**66**:2811.
170. Bultas MW, Steurer LM, Balakas K, Brooks C, Fields H. Psychosocial outcomes of a summer overnight recreational experience for children with heart disease. *Journal of Child Health Care* 2015;**19**:542-9. http://dx.doi.org/10.1177/1367493514540350

171. Desai P, Sutton L, Staley M, Hannon D. A qualitative study exploring the psychosocial value of weekend camping experiences for children and adolescents with complex heart defects. *Child Care Health Dev* 2014;**40**:553-61. http://dx.doi.org/http://dx.doi.org/10.1111/cch.12056
172. Gillard A, Allsop J. Camp experiences in the lives of adolescents with serious illnesses. *Children and Youth Services Review* 2016;**65**:112-9. http://dx.doi.org/10.1016/j.childyouth.2016.04.001
173. Hosek S, Brothers J, Lemos, the Adolescent Medicine Trials Network for HIV/AIDS Interventions D. What HIV-positive young women want from behavioral interventions: A qualitative approach. *AIDS Patient Care STDS* 2012;**26**:291-7.
174. Moola FJ, Faulkner G, White L, Kirsh J. Kids with special hearts: the experience of children with congenital heart disease at Camp Willowood. *Qualitiative Research in Sport, Exercise and Health* 2015;**7**:271-93. http://dx.doi.org/10.1080/2159676X.2014.926968
175. Stewart M, Barnfather A, Magill-Evans J, Ray L, Letourneau N. Brief report: an online support intervention: perceptions of adolescents with physical disabilities. *J Adolesc* 2011;**34**:795-800. http://dx.doi.org/http://dx.doi.org/10.1016/j.adolescence.2010.04.007
176. Barlow JH, Shaw KL, Harrison K. Consulting the 'experts': children's and parents' perceptions of psycho-educational interventions in the context of juvenile chronic arthritis. *Health Educ Res* 1999;**14**:597-610.
177. Fair CD, Connor L, Albright J, Wise E, Jones K. "I'm positive, I have something to say": Assessing the impact of a creative writing group for adolescents living with HIV. *The Arts in Psychotherapy* 2012;**39**:383-9.
178. Gillard A, Watts CE. Program features and developmental experiences at a camp for youth with cancer. *Children and Youth Services Review* 2013;**35**:890-8. http://dx.doi.org/10.1016/j.childyouth.2013.02.017
179. Gillard A, Witt PA, Watts CE. Outcomes and processes at a camp for youth with HIV/AIDS. *Qual Health Res* 2011;**21**:1508-26. http://dx.doi.org/10.1177/1049732311413907
180. Lewis P, Klineberg E, Towns S, Moore K, Steinbeck K. The Effects of Introducing Peer Support to Young People with a Chronic Illness. *Journal of Child and Family Studies* 2016;**25**:2541-53. http://dx.doi.org/10.1007/s10826-016-0427-4
181. Muskat B, Salter R, Shindler S, Porter M, Bitnun A. "Here you feel like it's not taboo": An evaluation of a pediatric hospital-based HIV support group. *Journal of HIV/AIDS & Social Services* 2016:1-18.
182. Nicholas DB, Darch J, McNeill T, Brister L, O'leary K, Berlin D*, et al.* Perceptions of online support for hospitalized children and adolescents. *Soc Work Health Care* 2007;**44**:205-23.
183. Stewart M, Letourneau N, Masuda JR, Anderson S, McGhan S. Online support for children with asthma and allergies. *J Fam Nurs* 2013;**19**:171-97. http://dx.doi.org/10.1177/1074840713483573
184. Tiemens K, Beveridge HL, Nicholas DB. Evaluation of a therapeutic camp program for adolescents with a facial difference. *Social Work with Groups* 2007;**30**:57-71.
185. White LC, Moola FJ, Kirsh JA, Faulkner GE. A Therapeutic Recreation Camp for Children with Congenital Heart Disease: Examining Impact on the Psychosocial Well-Being of Parents. *Journal of Child and Family Studies* 2016;**25**:3034-43.
186. Wright PM, White K, Gaebler-Spira D. Exploring the Relevance of the Personal and Social Responsibility Model in Adapted Physical Activity: A Collective Case Study. *Journal of Teaching in Physical Education* 2004;**23**:71-87.

187. Dennison L, Stanbrook R, Moss-Morris R, Yardley L, Chalder T. Cognitive behavioural therapy and psycho-education for chronic fatigue syndrome in young people: reflections from the families' perspective. *Br J Health Psychol* 2010;**15**:167-83. http://dx.doi.org/10.1348/135910709X440034
188. Kirk S, Milnes L. An exploration of how young people and parents use online support in the context of living with cystic fibrosis. *Health Expectations* 2016;**19**:309-21. http://dx.doi.org/10.1111/hex.12352
189. Serlachius A, Northam E, Frydenberg E, Cameron F. Adapting a generic coping skills programme for adolescents with type 1 diabetes: a qualitative study. *J Health Psychol* 2012;**17**:313-23. http://dx.doi.org/10.1177/1359105311415559
190. White L. *A Therapeutic Recreation Camp for Children with Congenital Heart Disease: Examining Impact on the Psycho-social Well-being of Caregivers and their Children*: University of Toronto; 2014.
191. Kashikar-Zuck S, Tran ST, Barnett K, Bromberg MH, Strotman D, Sil S*, et al.* A Qualitative Examination of a New Combined Cognitive-Behavioral and Neuromuscular Training Intervention for Juvenile Fibromyalgia. *Clin J Pain* 2016;**32**:70-81. http://dx.doi.org/10.1097/AJP.0000000000000221
192. Campbell T, Beer H, Wilkins R, Sherlock E, Merrett A, Griffiths J. "I look forward. I feel insecure but I am ok with it". The experience of young HIV+ people attending transition preparation events: a qualitative investigation. *AIDS Care* 2010;**22**:263-9. http://dx.doi.org/10.1080/09540120903111460
193. Curle C, Bradford J, Thompson J, Cawthron P. Users' views of a group therapy intervention for chronically Ill or disabled children and their parents: Towards a meaningful assessment of therapeutic effectiveness. *Clin Child Psychol Psychiatry* 2005;**10**:509-27. http://dx.doi.org/10.1177/1359104505056315
194. Gan C, Gargaro J, Kreutzer JS, Boschen KA, Wright FV. Development and preliminary evaluation of a structured family system intervention for adolescents with brain injury and their families. *Brain Inj* 2010;**24**:651-63. http://dx.doi.org/10.3109/02699051003692142
195. Jaser SS, Patel N, Linsky R, Whittemore R. Development of a positive psychology intervention to improve adherence in adolescents with type 1 diabetes. *J Pediatr Health Care* 2014;**28**:478-85. http://dx.doi.org/10.1016/j.pedhc.2014.02.008
196. Marsac ML, Hildenbrand AK, Clawson K, Jackson L, Kohser K, Barakat L*, et al.* Acceptability and feasibility of family use of The Cellie Cancer Coping Kit. *Support Care Cancer* 2012;**20**:3315-24. http://dx.doi.org/10.1007/s00520-012-1475-y
197. Masuda JR, Anderson S, Letourneau N, Sloan Morgan V, Stewart M. Reconciling Preferences and Constraints in Online Peer Support for Youth With Asthma and Allergies. *Health Promotion Practice* 2013;**14**:741-50. http://dx.doi.org/10.1177/1524839912465083
198. Nieto R, Hernandez E, Boixados M, Huguet A, Beneitez I, McGrath P. Testing the Feasibility of DARWeb: An Online Intervention for Children With Functional Abdominal Pain and Their Parents. *Clin J Pain* 2015;**31**:493-503. http://dx.doi.org/10.1097/AJP.0000000000000199
199. Nilsson S, Finnstrom B, Kokinsky E, Enskar K. The use of Virtual Reality for needle-related procedural pain and distress in children and adolescents in a paediatric oncology unit. *Eur J Oncol Nurs* 2009;**13**:102-9. http://dx.doi.org/10.1016/j.ejon.2009.01.003

200. Reme SE, Archer N, Chalder T. Experiences of young people who have undergone the Lightning Process to treat chronic fatigue syndrome/myalgic encephalomyelitis--a qualitative study. *Br J Health Psychol* 2013;**18**:508-25. http://dx.doi.org/10.1111/j.2044-8287.2012.02093.x
201. Sibinga EM, Kerrigan D, Stewart M, Johnson K, Magyari T, Ellen JM. Mindfulness-based stress reduction for urban youth. *J Altern Complement Med* 2011;**17**:213-8. http://dx.doi.org/10.1089/acm.2009.0605
202. Whittemore R, Grey M, Lindemann E, Ambrosino J, Jaser S. Development of an Internet coping skills training program for teenagers with type 1 diabetes. *Computers, informatics, nursing: CIN* 2010;**28**:103.
203. Wolf Bordonaro GP. Art therapy with hospitalized pediatric patients. *Dissertation Abstracts International Section A: Humanities and Social Sciences* 2005;**66**:1600.
204. Ayers S, Muller I, Mahoney L, Seddon P. Understanding needle-related distress in children with cystic fibrosis. *Br J Health Psychol* 2011;**16**:329-43. http://dx.doi.org/10.1348/135910710X506895
205. Barry P, O'Callaghan C, Wheeler G, Grocke D. Music therapy CD creation for initial pediatric radiation therapy: A mixed methods analysis. *J Music Ther* 2010;**47**:233-63.
206. Baruch JM. *The Beads of Courage program for children coping with cancer*: University of Arizona; 2010.
207. Burns DS, Robb SL, Phillips-Salimi C, Haase JE. Parental perspectives of an adolescent/young adult stem cell transplant and a music video intervention. *Cancer Nurs* 2010;**33**:E20-7. http://dx.doi.org/10.1097/NCC.0b013e3181d4b671
208. Docherty SL, Robb SL, Phillips-Salimi C, Cherven B, Stegenga K, Hendricks-Ferguson V*, et al.* Parental Perspectives on a Behavioral Health Music Intervention for Adolescent/Young Adult Resilience During Cancer Treatment: Report From the Children's Oncology Group. *J Adolesc Health* 2013;**52**:170-8. http://dx.doi.org/10.1016/j.jadohealth.2012.05.010
209. O'Callaghan C, Baron A, Barry P, Dun B. Music's relevance for pediatric cancer patients: a constructivist and mosaic research approach. *Support Care Cancer* 2011;**19**:779-88. http://dx.doi.org/10.1007/s00520-010-0879-9
210. O'Callaghan C, Barry P, Thompson K. Music's relevance for adolescents and young adults with cancer: a constructivist research approach. *Support Care Cancer* 2012;**20**:687-97. http://dx.doi.org/10.1007/s00520-011-1104-1
211. O'Callaghan C, Dun B, Baron A, Barry P. Music's relevance for children with cancer: music therapists' qualitative clinical data-mining research. *Soc Work Health Care* 2013;**52**:125-43. http://dx.doi.org/10.1080/00981389.2012.737904
212. Shrimpton BJ, Willis DJ, Tongs CD, Rolfo AG. Movie making as a cognitive distraction for paediatric patients receiving radiotherapy treatment: qualitative interview study. *BMJ Open* 2013;**3**. http://dx.doi.org/10.1136/bmjopen-2012-001666
213. Weekes DP, Kagan SH, James K, Seboni N. The phenomenon of hand holding as a coping strategy in adolescents experiencing treatment-related pain. *J Pediatr Oncol Nurs* 1993;**10**:19-25.
214. Barnfather A, Stewart M, Magill-Evans J, Ray L, Letourneau N. Computer-mediated support for adolescents with cerebral palsy or spina bifida. *CIN: Computers, Informatics, Nursing* 2011;**29**:24-35 12p. http://dx.doi.org/10.1097/NCN.0b013e3181f9db63
215. Brothers J, Harper GW, Fernandez M, Hosek SG. EVOLUTION-Taking charge and growing stronger: The design, acceptability, and feasibility of a secondary prevention

empowerment intervention for young women living with HIV. *AIDS Patient Care STDS* 2014;**28**:33-42. http://dx.doi.org/10.1089/apc.2013.0085
216. Gaysynsky A, Romansky-Poulin K, Arpadi S. "My YAP Family": Analysis of a Facebook Group for Young Adults Living with HIV. *AIDS Behav* 2015;**19**:947-62. http://dx.doi.org/10.1007/s10461-014-0887-8
217. Griffiths C, Panteli N, Brunton D, Marder B, Williamson H. Designing and evaluating the acceptability of Realshare: An online support community for teenagers and young adults with cancer. *J Health Psychol* 2015;**20**:1589-601. http://dx.doi.org/10.1177/1359105313519154
218. MacDonald K, Greggans A. 'Cool friends': an evaluation of a community befriending programme for young people with cystic fibrosis. *J Clin Nurs* 2010;**19**:2406-14.
219. Nicholas DB, Fellner KD, Frank M, Small M, Hetherington R, Slater R*, et al.* Evaluation of an online education and support intervention for adolescents with diabetes. *Soc Work Health Care* 2012;**51**:815-27.
220. Nicholas DB, Williams M, MacLusky IB. Evaluating group work within a summer camp intervention for children and adolescents with asthma. *Social Work with Groups* 2009;**32**:209-21.
221. Romero SL. The development and impact of an online social networking community for adolescents with cystic fibrosis. *Dissertation Abstracts International: Section B: The Sciences and Engineering* 2014;**74**:No Pagination Specified.
222. Stewart M, Letourneau N, Masuda JR, Anderson S, McGhan S. Impacts of online peer support for children with asthma and allergies: It just helps you every time you can't breathe well". *J Pediatr Nurs* 2013;**28**:439-52. http://dx.doi.org/10.1016/j.pedn.2013.01.003
223. Stewart M, Masuda JR, Letourneau N, Anderson S, McGhan S. "I want to meet other kids like me": support needs of children with asthma and allergies. *Issues Compr Pediatr Nurs* 2011;**34**:62-78. http://dx.doi.org/10.3109/01460862.2011.572638
224. Stinson J, McGrath P, Hodnett E, Feldman B, Duffy C, Huber A*, et al.* Usability testing of an online self-management program for adolescents with juvenile idiopathic arthritis. *J Med Internet Res* 2010;**12**:e30.
225. Stinson JN, Toomey PC, Stevens BJ, Kagan S, Duffy CM, Huber A*, et al.* Asking the experts: Exploring the self-management needs of adolescents with arthritis. *Arthritis Care Res* 2008;**59**:65-72.
226. Atkins S, Lewin S, Smith H, Engel M, Fretheim A, Volmink J. Conducting a meta-ethnography of qualitative literature: lessons learnt. *BMC Med Res Methodol* 2008;**8**:21.
227. Waytz A, Gray K, Epley N, Wegner DM. Causes and consequences of mind perception. *Trends in cognitive sciences* 2010;**14**:383-8.
228. Cutrona CE, Suhr JA. Controllability of stressful events and satisfaction with spouse support behaviors. *Communication Research* 1992;**19**:154-74.
229. Health JCPfM. *Guidance for commissioners of child and adolescent mental health services*; 2013.
230. Thorley C. *Education, Education, Mental Health*. London: Institute for Public Policy Research; 2016.
231. Minds Y. *Mental health statistics*. 2017. URL: http://www.youngminds.org.uk/training_services/policy/mental_health_statistics (Accessed 10th April 2017).
232. Taskforce MH. *The five year forward view for mental health*. Redditch: National Health Service; 2016.

233. Choices N. *Carers' breaks and respite care*. 2015. URL: http://www.nhs.uk/Conditions/social-care-and-support-guide/Pages/breaks-for-carers-respite-care.aspx (Accessed 21st March 2017).
234. Richards DA, Hallberg IR. *Complex interventions in health: an overview of research methods*: Routledge; 2015.
235. Sandelowski M, Voils CI, Barroso J. Defining and designing mixed research synthesis studies. *Research in the schools: a nationally refereed journal sponsored by the Mid-South Educational Research Association and the University of Alabama* 2006;**13**:29.
236. Whitaker R, Hendry M, Rabeea'h Aslam AB, Carter B, Charles JM, Craine N*, et al.* Intervention Now to Eliminate Repeat Unintended Pregnancy in Teenagers (INTERUPT): a systematic review of intervention effectiveness and cost-effectiveness, and qualitative and realist synthesis of implementation factors and user engagement. *Health technology assessment (Winchester, England)* 2016;**20**:1.
237. Noyes J, Popay J. Directly observed therapy and tuberculosis: how can a systematic review of qualitative research contribute to improving services? A qualitative meta-synthesis. *J Adv Nurs* 2007;**57**:227-43.
238. Glenton C, Colvin CJ, Carlsen B, Swartz A, Lewin S, Noyes J*, et al.* Barriers and facilitators to the implementation of lay health worker programmes to improve access to maternal and child health: qualitative evidence synthesis. *The Cochrane Library* 2013.
239. Thomas J, Harden A, Oakley A, Oliver S, Sutcliffe K, Rees R*, et al.* Integrating qualitative research with trials in systematic reviews. *BMJ: British Medical Journal* 2004;**328**:1010.
240. Harn B, Parisi D, Stoolmiller M. Balancing fidelity with flexibility and fit: What do we really know about fidelity of implementation in schools? *Except Child* 2013;**79**:181-93.
241. Frydenberg* E, Lewis R, Bugalski K, Cotta A, McCarthy C, Luscombe-smith N*, et al.* Prevention is better than cure: Coping skills training for adolescents at school. *Educational Psychology in Practice* 2004;**20**:117-34.
242. Beresford P. User involvement, research and health inequalities: developing new directions. *Health & Social Care in the Community* 2007;**15**:306-12.
243. Boote J, Baird W, Beecroft C. Public involvement at the design stage of primary health research: a narrative review of case examples. *Health Policy* 2010;**95**:10-23.
244. Thompson J, Barber R, Ward PR, Boote JD, Cooper CL, Armitage CJ*, et al.* Health researchers' attitudes towards public involvement in health research. *Health Expectations* 2009;**12**:209-20.
245. Thompson-Coon J, Gwernan-Jones R, Moore D, Richardson M, Shotton C, Pritchard W*, et al.* End-user involvement in a systematic review of quantitative and qualitative research of non-pharmacological interventions for attention deficit hyperactivity disorder delivered in school settings: reflections on the impacts and challenges. *Health Expectations* 2015; 10.1111/hex.12400:n/a-n/a. http://dx.doi.org/10.1111/hex.12400
246. Mockford C, Staniszewska S, Griffiths F, Herron-Marx S. The impact of patient and public involvement on UK NHS health care: a systematic review. *Int J Qual Health Care* 2011;**24**:28-38.
247. Moore GF, Audrey S, Barker M, Bond L, Bonell C, Hardeman W*, et al.* Process evaluation of complex interventions: Medical Research Council guidance. *BMJ* 2015;**350**:h1258.
248. Schulz KF, Altman DG, Moher D. CONSORT 2010 statement: updated guidelines for reporting parallel group randomised trials. *BMC Med* 2010;**8**:18.

249. Foundation K. *Logic model development guide*. Battle Creek, MI: Kellogg Foundation; 2001.
250. Williamson PR, Altman DG, Blazeby JM, Clarke M, Gargon E. The COMET (core outcome measures in effectiveness trials) initiative. *Trials* 2011;**12**:A70.
251. Slaski M, Cartwright S. Emotional intelligence training and its implications for stress, health and performance. *Stress and health* 2003;**19**:233-9.

Appendix 1: Search strategy used for Review 1 with the Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) database

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1 Chronic Disease/px [Psychology] (4269)

2 Neoplasms/px [Psychology] (15475)

3 Diabetes Mellitus/px [Psychology] (2700)

4 Asthma/px [Psychology] (3457)

5 exp Respiratory Tract Diseases/px [Psychology] (12446)

6 Cystic Fibrosis/px [Psychology] (854)

7 Cerebral Palsy/px [Psychology] (1058)

8 exp Epilepsy/px [Psychology] (7330)

9 exp Muscular Diseases/px [Psychology] (4743)

10 exp Endocrine System Diseases/px [Psychology] (14089)

11 exp Immune System Diseases/px [Psychology] (33503)

12 exp HIV Infections/px [Psychology] (16557)

13 exp Cardiovascular Diseases/px [Psychology] (27191)

14 exp Nervous System Diseases/px [Psychology] (128905)

15 exp Skin Diseases/px [Psychology] (14730)

16 exp Digestive System Diseases/px [Psychology] (11110)

17 exp Hematologic Diseases/px [Psychology] (2320)

18 exp Otorhinolaryngologic Diseases/px [Psychology] (6693)

19 exp Stomatognathic Diseases/px [Psychology] (5416)

20 exp Eye Diseases/px [Psychology] (4290)

21 exp Pain/px [Psychology] (23130)

22 Disabled Children/px [Psychology] (941)

23 ((chronic* or longterm or long-term or "long standing" or physical) adj2 (condition* or ill* or disease* or disorder* or syndrome*)).ti,ab. (204183)

24 (cancer* or neoplas* or tumor* or tumour* or malignan* or carcinoma* or "bone marrow" or leukaemia or leukemia).ti,ab. (2710274)

25 “spina bifada” .ti,ab. (458788)

26 asthma*.ti,ab. (127105)

27 (respiratory adj2 (illness* or disease* or condition*)).ti,ab. (32642)

28 cystic fibrosis.ti,ab. (35054)

29 cerebral palsy.ti,ab. (16321)

30 quadriplegi*.ti,ab. (3604)

31 tetraplegi*.ti,ab. (3450)

32 diplegi*.ti,ab. (1995)

33 spinal cord injur*.ti,ab. (27628)

34 muscular dystrophy.ti,ab. (17776)

35 epilep*.ti,ab. (105645)

36 seizure*.ti,ab. (93277)

37 spina bifida.ti,ab. (5893)

38 (heart adj2 (condition* or disease* or disorder* or defect*)).ti,ab. (147955)

39 (cardiac adj2 (condition* or disease* or disorder* or defect*)).ti,ab. (27109)

40 (cardiovascular adj2 (condition* or disease* or disorder* or defect*)).ti,ab. (121138)

41 (skin adj2 (condition* or disease* or disorder*)).ti,ab. (27379)

42 eczema.ti,ab. (13093)

43 (allergies or allergy).ti,ab. (60164)

44 dermatitis.ti,ab. (44017)

45 (gastrointestinal adj (disorder* or disease*)).ti,ab. (11476)

46 ((stomach or abdominal or gastrointestinal) adj pain).ti,ab. (41843)

47 (bowel* adj2 inflammatory adj2 (condition* or disease* or illness*)).ti,ab. (31340)

48 (liver adj (disease* or transplant*)).ti,ab. (104995)

49 hepatitis.ti,ab. (175155)

50 (disabilit* adj5 child*).ti,ab. (7935)

51 (human immunodeficiency virus or HIV).ti,ab. (262555)

52 AIDS.ti,ab. (127472)

53 (hyperthyroidism or hypothyroidism).ti,ab. (38333)

54 an?emia.ti,ab. (113434)

55 h?emophilia.ti,ab. (15632)

56 sickle.ti,ab. (20240)

57 ((renal or kidney) adj (disease* or disorder*)).ti,ab. (87486)

58 nephrotic syndrome.ti,ab. (15366)

59 encephalomyelitis.ti,ab. (15778)

60 chronic fatigue syndrome.ti,ab. (4389)

61 ((cleft or palate) adj lip).ti,ab. (10349)

62 craniofacial.ti,ab. (17695)

63 (deaf or deafness).ti,ab. (24812)

64 (hearing adj (defect* or disorder*)).ti,ab. (1915)

65 blindness.ti,ab. (20927)

66 ((vision or visually or visual) adj (impaired or impairment*)).ti,ab. (9360)

67 ((persistent or chronic or recurring or frequent) adj (headache* or migraine*)).ti,ab. (3532)

68 chronic pain.ti,ab. (23794)

69 fibromyalgia.ti,ab. (7465)

70 medically unexplained symptoms.ti,ab. (505)

71 (spinal adj injur*).ti,ab. (4324)

72 or/1-71 (4906406)

73 (child or children*).ti,ab. (959407)

74 (adolescent or adolescents).ti,ab. (174139)

75 teen*.ti,ab. (23472)

76 (young adj (adult* or people)).ti,ab. (81908)

77 youth*.ti,ab. (46618)

78 73 or 74 or 75 or 76 or 77 (1146540)

79 exp Mental Disorders/pc, rh, th [Prevention & Control, Rehabilitation, Therapy] (219183)

80 exp Self-Injurious Behavior/pc, th [Prevention & Control, Therapy] (10462)

81 (mental adj (health or illness* or disorder* or distress or problem*)).ti,ab. (123672)

82 (psychiatric adj (illness* or disorder* or difficulties or problems or distress or adjustment)).ti,ab. (37713)

83 (psychological adj (illness* or disorder* or difficulties or problems or distress)).ti,ab. (18705)

84 psychopathology.ti,ab. (23584)

85 (depression or depressed or depressive).ti,ab. (321415)

86 adjustment disorders.ti,ab. (578)

87 (anxiety or anxious).ti,ab. (131581)

88 (behavio*ral adj (condition* or disturb* or disorder*)).ti,ab. (7142)

89 mood disorder*.ti,ab. (12037)

90 (emotional adj (disorder* or distress or adjustment or problem* or health)).ti,ab. (11716)

91 psychosocial adjustment.ti,ab. (1720)

92 internali?ing.ti,ab. (5832)

93 (self adj (harm* or injur* or mutilation)).ti,ab. (7223)

94 bipolar.ti,ab. (47112)

95 conduct disorder*.ti,ab. (3658)

96 externali?ing.ti,ab. (5649)

97 oppositional defiant.ti,ab. (1599)

98 ((disruptive or challenging or antisocial) adj behavio?r).ti,ab. (5296)

99 eating disorder*.ti,ab. (13689)

100 anorexia.ti,ab. (23853)

101 bulimia.ti,ab. (6163)

102 mutism.ti,ab. (1712)

103 obsess*.ti,ab. (15553)

104 panic disorder*.ti,ab. (8123)

105 phobia*.ti,ab. (7304)

106 post traumatic stress disorder.ti,ab. (6641)

107 PTSD.ti,ab. (14726)

108 (psychosis or psychotic).ti,ab. (44807)

109 personality disorder*.ti,ab. (15416)

110 mania.ti,ab. (8238)

111 schizophrenia.ti,ab. (83194)

112 (self harm or self injur*).ti,ab. (5930)

113 (suicide or suicidal).ti,ab. (54111)

114 or/79-113 (896398)

115 (substance adj (misuse or abuse)).ti,ab. (20418)

116 ((drug or alcohol) adj (addiction or abuse or misuse)).ti,ab. (33943)

117 115 or 116 (51906)

118 12 or 49 or 51 or 52 (486920)

119 117 not 118 (46257)

120 114 or 119 (917885)

121 randomized controlled trial.pt. (404549)

122 controlled clinical trial.pt. (90003)

123 exp Randomized Controlled Trials as Topic/ (101462)

124 randomi?ed.ti,ab. (426544)

125 randomly.ab. (240673)

126 trial.ti,ab. (407141)

127 groups.ab. (1507035)

128 or/121-127 (2247880)

129 72 and 78 and 120 and 128 (4473)

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Appendix 2: Search strategy used for Review 1 economic search with the Ovid EMBASE database

1 exp childhood disease/cn, dm, rh, th [Congenital Disorder, Disease Management, Rehabilitation, Therapy] 10053

2 diabetes mellitus/rh, th [Rehabilitation, Therapy] 13071

3 asthma/rh, th [Rehabilitation, Therapy] 10837

4 respiratory tract disease/rh, th [Rehabilitation, Therapy] 2064

5 cystic fibrosis/dm, rh, th [Rehabilitation, Therapy] 3304

6 cerebral palsy/dm, rh, th [Rehabilitation, Therapy] 4379

7 epilepsy/rh, th [Rehabilitation, Therapy] 5014

8 heart disease/dm, rh, th [Rehabilitation, Therapy] 4816

9 skin disease/rh, th [Rehabilitation, Therapy] 3943

10 muscle disease/rh, th [Rehabilitation, Therapy] 838

11 endocrine disease/rh, th [Rehabilitation, Therapy] 428

12 neurologic disease/rh, th [Rehabilitation, Therapy] 4281

13 hematologic disease/rh, th [Rehabilitation, Therapy] 1991

14 mouth disease/rh, th [Rehabilitation, Therapy] 1958

15 ear disease/rh, th [Rehabilitation, Therapy] 341

16 eye disease/rh, th [Rehabilitation, Therapy] 2159

17 handicapped child/ 7141

18 ((chronic* or longterm or long-term or "long standing" or physical) adj2 (condition* or ill* or disease* or disorder* or syndrome*)).ti,ab. 295589

19 (cancer* or neoplas* or tumor* or tumour* or malignan* or carcinoma* or "bone marrow" or leukaemia or leukemia).ti,ab. 3623580

20 diabet*.ti,ab. 684993

21 asthma*.ti,ab. 185088

22 (respiratory adj2 (illness* or disease* or condition*)).ti,ab. 44620

23 cystic fibrosis.ti,ab. 50040

24 cerebral palsy.ti,ab. 23043

25 quadriplegi*.ti,ab. 4721

26 tetraplegi*.ti,ab. 4658

27 diplegi*.ti,ab. 2764

28 spinal cord injur*.ti,ab. 36614

29 muscular dystrophy.ti,ab. 22965

30 epilep*.ti,ab. 157765

31 seizure*.ti,ab. 140681

32 spina bifida.ti,ab. 7573

33 (heart adj2 (condition* or disease* or disorder* or defect*)).ti,ab. 204821

34 (cardiac adj2 (condition* or disease* or disorder* or defect*)).ti,ab. 39924

35 (cardiovascular adj2 (condition* or disease* or disorder* or defect*)).ti,ab. 178430

36 (skin adj2 (condition* or disease* or disorder*)).ti,ab. 40227

37 eczema.ti,ab. 19576

38 (allergies or allergy).ti,ab. 94945

39 dermatitis.ti,ab. 62293

40 (gastrointestinal adj (disorder* or disease*)).ti,ab. 16480

41 ((stomach or abdominal or gastrointestinal) adj pain).ti,ab. 67844

42 (bowel* adj2 inflammatory adj2 (condition* or disease* or illness*)).ti,ab. 50014

43 (liver adj (disease* or transplant*)).ti,ab. 162025

44 hepatitis.ti,ab. 238818

45 (disabilit* adj5 child*).ti,ab. 10810

46 (human immunodeficiency virus or HIV).ti,ab. 325933

47 AIDS.ti,ab. 146763

48 (hyperthyroidism or hypothyroidism).ti,ab. 51321

49 an?emia.ti,ab. 160099

50 h?emophilia.ti,ab. 24268

51 sickle.ti,ab. 27249

52 ((renal or kidney) adj (disease* or disorder*)).ti,ab. 125441

53 nephrotic syndrome.ti,ab. 19220

54 encephalomyelitis.ti,ab. 19754

55 chronic fatigue syndrome.ti,ab. 5809

56 ((cleft or palate) adj lip).ti,ab. 11586

57 craniofacial.ti,ab. 21025

58 (deaf or deafness).ti,ab. 30007

59 (hearing adj (defect* or disorder*)).ti,ab. 2285

60 blindness.ti,ab. 26167

61 ((vision or visually or visual) adj (impaired or impairment*)).ti,ab. 13041

62 ((persistent or chronic or recurring or frequent) adj (headache* or migraine*)).ti,ab. 6194

63 chronic pain.ti,ab. 36382

64 fibromyalgia.ti,ab. 12217

65 medically unexplained symptoms.ti,ab. 666

66 (spinal adj injur*).ti,ab. 5422

67 or/1-66 6477448

68 (child or children*).ti,ab. 1248757

69 (adolescent or adolescents).ti,ab. 232830

70 teen*.ti,ab. 31160

71 (young adj (adult* or people)).ti,ab. 109824

72 youth*.ti,ab. 58783

73 68 or 69 or 70 or 71 or 72 1487904

74 mental disease/dm, pc, rh, th [Disease Management, Prevention, Rehabilitation, Therapy] 30502

75 automutilation/dm, pc, rh, th [Disease Management, Prevention, Rehabilitation, Therapy] 1490

76 depression/dm, pc, rh, th [Disease Management, Prevention, Rehabilitation, Therapy] 28465

77 exp anxiety disorder/dm, rh, th [Disease Management, Rehabilitation, Therapy] 22583

78 exp eating disorder/dm, pc, rh, th [Disease Management, Prevention, Rehabilitation, Therapy] 6782

79 (mental* adj2 (ill* or disorder*)).ti,ab. 71484

80 (psychiatric adj (illness* or disorder* or difficulties or problems or distress or adjustment)).ti,ab. 55442

81 (psychological adj (illness* or disorder* or difficulties or problems or distress)).ti,ab. 25749

82 psychopathology.ti,ab. 32344

83 (depression or depressed or depressive).ti,ab. 440151

84 adjustment disorders.ti,ab. 904

85 (anxiety or anxious).ti,ab. 193926

86 (behavio*ral adj (condition* or disturb* or disorder*)).ti,ab. 10574

87 mood disorder*.ti,ab. 18825

88 (emotional adj (disorder* or distress or adjustment or problem* or health)).ti,ab. 16428

89 psychosocial adjustment.ti,ab. 2176

90 internali?ing.ti,ab. 7243

91 (self adj (harm* or injur* or mutilation)).ti,ab. 9543

92 bipolar.ti,ab. 68758

93 conduct disorder*.ti,ab. 4850

94 externali?ing.ti,ab. 7009

95 oppositional defiant.ti,ab. 2086

96 ((disruptive or challenging or antisocial) adj behavio?r).ti,ab. 6916

97 eating disorder*.ti,ab. 19058

98 anorexia.ti,ab. 33225

99 bulimia.ti,ab. 8250

100 mutism.ti,ab. 2429

101 obsess*.ti,ab. 22459

102 panic disorder*.ti,ab. 10807

103 phobia*.ti,ab. 10326

104 post traumatic stress disorder.ti,ab. 9149

105 PTSD.ti,ab. 20214

106 (psychosis or psychotic).ti,ab. 69543

107 personality disorder*.ti,ab. 21402

108 mania.ti,ab. 12209

109 schizophrenia.ti,ab. 118359

110 (self harm or self injur*).ti,ab. 7961

111 (suicide or suicidal).ti,ab. 70596

112 (substance adj (misuse or abuse)).ti,ab. 27665

113 ((drug or alcohol) adj (addiction or abuse or misuse)).ti,ab. 47759

114 or/74-111 998337

115 112 or 113 72030

116 44 or 46 or 47 618316

117 115 not 116 64343

118 114 or 117 1041237

119 67 and 73 and 118 23307

120 health economics/ 35400

121 exp Economic Evaluation/ 241708

122 exp Health Care Cost/ 232673

123 pharmacoeconomics/ 6278

124 (econom* or cost or costs or costly or costing or price or prices or pricing or pharmacoeconomic*).ti,ab. 733411

125 (expenditure$ not energy).ti,ab. 28398

126 (value adj2 money).ti,ab. 1693

127 budget*.ti,ab. 28298

128 120 or 121 or 122 or 123 or 124 or 125 or 126 or 127 959359

129 119 and 128 1258

130 limit 129 to yr="2015 -Current" 129

Appendix 3: List of websites searched

Mental Health Foundation https://www.mentalhealth.org.uk/

Young Minds http://www.youngminds.org.uk/

Department of Child and Adolescent Mental Health Service (CAMHS) at Great Ormond Street Hospital http://www.gosh.nhs.uk/medical-information/clinical-specialties/child-and-adolescent-mental-health-services-camhs-information-parents-and-visitors/research-and-publications

Child & Adolescent Mental Health Services (CAMHS) Research Unit http://www.cpcs.org.uk/index.php?page=high-needs-project

NIHR CRN: Children and young people's mental health research https://www.crn.nihr.ac.uk/mentalhealth/about-mental-health-research/children-and-young-peoples-mental-health-research/

NIMH Child and Adolescent Mental Health http://www.nimh.nih.gov/health/topics/child-and-adolescent-mental-health/index.shtml#part_152583

RCP CAMHS Resource Library http://www.rcpsych.ac.uk/quality/qualityandaccreditation/childandadolescent/communitycamhsqncc/camhsresourcelibrary.aspx

National Child and Maternal Health Intelligence Network http://www.chimat.org.uk/camhs

National Children's Bureau http://www.ncb.org.uk/healthy-care/useful-links/mental-health-and-emotional-well-being

Association for Children's Mental Health http://www.acmh-mi.org/get-information/

Substance Abuse and Mental Health Services Administration http://www.samhsa.gov/data/

American Academy of Pediatrics https://www.aap.org/en-us/professional-resources/Research/Pages/Research.aspx

Child and Youth Health http://www.cyh.com/HealthTopics/HealthTopicDetails.aspx

The Kings Fund: Mental Health http://www.kingsfund.org.uk/topics/mental-health

Additions found by Liz:

Young Epilepsy: http://www.youngepilepsy.org.uk/

Epilepsy Society: https://www.epilepsysociety.org.uk/young-people-and-epilepsy#.Vz8FjuTAN_A

Epilepsy Action: https://www.epilepsy.org.uk/info/young-people

Cancer Research UK: http://www.cancerresearchuk.org/health-professional/cancer-statistics/teenagers-and-young-adults-cancers

Macmillan: http://www.macmillan.org.uk/cancerinformation/teensandyoungadults/infoforteensandyoungadults.aspx

Teenage Cancer Trust: https://www.teenagecancertrust.org/get-help/ive-got-cancer/stories-others

Teenagers and Young Adults with Cancer: http://www.tyac.org.uk/

Sickle Cell and young stroke survivors: http://www.scyss.org/

Sickle Cell Society: http://sicklecellsociety.org/resources/did-you-know-age-11-16-an-information-booklet-for-young-people/

AVERT (HIV): http://www.avert.org/living-with-hiv/health-wellbeing/being-young-positive

Young people with Aids: http://www.amfar.org/about-hiv-and-aids/young-people-and-hiv/young-people-and-hiv-aids/

Pozitude: http://www.pozitude.co.uk/

Children and Young People HIV network: http://www.ncb.org.uk/hiv

Child's Brain Injury Trust: http://childbraininjurytrust.org.uk/how-we-help/young-people/

Stroke Association: https://www.stroke.org.uk/finding-support/information-about-stroke-childhood

National Institute of Neurological Disorders and Stroke: http://www.ninds.nih.gov/disorders/tbi/detail_tbi.htm

Cerbera: http://w3.cerebra.org.uk/

Association of young people with M.E.: http://www.ayme.org.uk/

Away with Pain: http://www.awaywithpain.co.uk/young-people

Fibromyalgia: http://www.fmauk.org/information-packs-mainmenu-58/booklet-mainmenu-135/232-children-and-young-people

http://www.fmscommunity.org/pediatric.htm

Healing Well: http://www.healingwell.com/community/default.aspx?f=24&m=1813258

Arthritis research UK: http://www.arthritisresearchuk.org/arthritis-information/young-people.aspx

Youth Health: http://www.healthtalk.org/young-peoples-experiences/arthritis/what-arthritis-and-what-are-early-symptoms

Arthritis Care: https://www.arthritiscare.org.uk/managing-arthritis/children-with-arthritis

CLIC Sargent http://www.clicsargent.org.uk/content/help-and-support

Cerebral Palsy . org http://www.cerebralpalsy.org/information/child-care

Asthma UK https://www.asthma.org.uk/research/

British Lung Foundation https://www.blf.org.uk/what-we-do/research

Diabetes UK https://www.diabetes.org.uk/Research/

British Deaf Association https://www.bda.org.uk/publications

Mental Elf http://www.nationalelfservice.net/

Appendix 4: Categories of intervention, their definition and included studies which use them.

Intervention	Definition	Studies
Cognitive Behavioural Therapy (CBT)	Structured programme which aims to identify negative thought processes, which may be triggered by certain situations or physical feelings, and learn to think and act upon them in a positive way. (http://www.nhs.uk/Conditions/Cognitive-behavioural-therapy/Pages/How-does-it-work.aspx)	82, 83, 123-125, 129, 145
Parenting Programmes	Family interventions aiming to reduce child behavioural and emotional difficulties by targeting improvement in parenting skills, knowledge and confidence (from Brown et al., 2014[127])	117, 130
Parenting Programmes with Acceptance and Commitment Therapy	As above, but with the addition of acceptance and commitment therapy (ACT). ACT is a variation of CBT incorporating mindfulness techniques which aim to increase psychological flexibility and decrease experiential avoidance (from Brown et al., 2014[127])	127, 130
Palliative care	Interventions which focus on making the patient as comfortable as possible by helping to manage pain and symptoms, and provide psychological, social and spiritual support for the patient, their family or carers. Commonly thought of as end of life care, but this is not always true. (http://www.nhs.uk/Planners/end-of-life-care/Pages/what-it-involves-and-when-it-starts.aspx)	136, 140

Intervention	Definition	Studies
Group Play Therapy	Interventions where children and therapists use games and play activities as the main medium of communication, with the aim of achieving a psychological interaction that conveys therapeutic benefit (from Wang et al., 2012[137])	132, 134, 137
Emotional intelligence training	Programmes designed to improve emotional intelligence, which is the skills with which to perceive and regulate emotions, particularly with regard to the development of stress. (Slaski & Cartwright, 2003[251])	131, 133
Relaxation	Interventions using one or more generic techniques for relaxation, including breathing exercises, progressive muscle relaxation or guided imagery, to reduce psychological distress.	118, 138
Stress management training	A structured programme aiming to improve coping with stress. While aspects of other interventions may be used (e.g. CBT/cognitive restructuring), they form one of an array of coping strategies. Other stress management techniques may involve goal setting, medical adherence, emotional control etc.	119, 121
Music therapy	Interventions where music is delivered by therapists in a way that provides psychosocial support. Involves creating music with instruments or voice, and is distinct from passively listening to music. (http://www.bamt.org/)	135

Intervention	Definition	Studies
Massage therapy	Manual manipulation of soft body tissues (muscle, connective tissue, tendons and ligaments) to enhance a person's health and well-being. (https://www.nwhealth.edu/school-of-massage-therapy/massage-therapy-definition/)	[120]
Heart Rate Variability Biofeedback	Combines the use of relaxation techniques with non-invasive technology providing real-time feedback on heart rate. (from Yetwin)	[126]
Resistance Training	A graded exercise programme involving sets of lifting, pushing or pulling activities (including free weights/objects, resistance training machines or participant bodyweight).	[128]

Appendix 5: Categories of intervention, and details of delivery and adapted components.

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
CBT (CBI)	Martinovic (2006)[82]	No	No manual. Delivered 'as part of individual treatment plan' – but no details.	Individual treatment plan aimed at distorted automatic thoughts related to negative depressive thinking.
CBT (TAPS)	Masia Warner (2011)[123]	Yes	Modified from 'Coping Cat'. Option to receive CBT at one of two locations.	Jointly addresses anxiety and physical symptoms. Identifies contexts in which symptoms occur, targets fears related to physical pain and anxiety-inducing situations.
CBT (TAPS+IBD)	Reigada (2015)[124]	Yes	Adapted from 'TAPS'. Delivered to all from a treatment manual.	Specifically focuses on interaction of anxiety and IBD symptoms. Included content designed to assist CYP with academic and social activities when physical symptoms present, as well as IBD management and reacting positively to pain. Parents encouraged to foster independence in the CYP.
CBT (BOC)	Serlachius (2014)[129]	Yes	Based on manualised 'Best of Coping', with added components. Not explicit, but diaries and goals likely individualised.	Diabetes-specific components were added to the standard Best Of Coping intervention to specifically address the needs of CYP with T1DM. Added components: conflict resolution; health-behaviour diaries to support T1DM management and goals; positive reinforcement and modelling to increase T1DM-related self-efficacy; T1DM-specific information and resources.

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
CBT (PASCET-PI)	Szigethy (2007)[83]	Yes	Based on PASCET. Allowed telephone delivery.	Adaptation of PASCET, with addition of physical illness narrative. Targets depression, but has specific content related to IBD to improve cognitions and behaviours related to IBD. Includes information about IBD, techniques for coping with abdominal pain, identifying negative cognitions about IBD, eliciting and later reassessing the CYP's physical illness narrative (belief about how IBD influences their life).
CBT (PASCET-PI)	Szigethy (2014)[125]	Yes	Content was simplified or made more child-friendly for younger participants (age 9-13). More telephone delivery than 2007 version – allowed greater flexibility for participants.	As above, but specifically states that the programme was "tailored to a youth's individual developmental stage". This appears to be manifested in simplifying materials for 9-13 y/o – "For participants aged 9 to 13 years receiving CBT, handouts and practice assignments were simplified; pictures were used to illustrate concepts; skills became part of games; and parents were involved after each individual session to review practice assignments. In SNDT, conversations with participants aged 9 to 13 years were conducted during games, and parents were involved after each session to review their child's progress". They also considered the delivery of CBT over the phone where possible, as a response to the wide catchment area of participating families.

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
CBT (ACT)	Wicksell (2009)[145]	Yes	Participants received intervention individually. Unclear whether all received exactly the same content – no manual.	Entire intervention focused on changing CYP's response to chronic pain. Included thinking about pain differently, trying to accept it but not let it limit activities, and thus reduce avoidant behaviours.
Parenting Programme (SSTP)	Whittingham (2014)[130]	No	Based on SSTP manual	Parenting skills for CYP with behavioural problems and a disability. No mention of content specifically related to LTC (Cerebral Palsy).
Parenting Programme (Triple P)	Westrupp (2015)[117]	No	Based on Triple P manual	Standard Triple P. No diabetes-specific content.
Parenting Programme with Acceptance & Commitment Therapy (SSTP+ACT)	Whittingham (2014)[130]	No	Based on SSTP manual, but with added ACT.	As with SSTP (Whittingham 2014), but with additional ACT exercises designed to improve parent coping with stress and improvement in psychological skills. ACT contains no description of any content specifically related to having a CYP with an LTC (Cerebral Palsy).

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
Parenting Programme with Acceptance & Commitment Therapy (SSTP+ACT)	Brown (2014)[127]	No	Based on SSTP manual, but with added ACT.	No content was adapted specifically to the LTC (acquired brain injury).
Palliative care (ACP)	Lyon (2013)[140]	No	Structured delivery based on Lyon Family-Centred ACP. Five Wishes exercise was part of shared decision making for under 18s.	Only LTC-specific component was increasing understanding of the medical condition. In this study it was cancer, but could be any condition. Remaining content not LTC-specific.
Palliative care (Make a Wish)	Shoshani (2015)[136]	No	All CYP interviewed at home. No variation in interview.	No suggestion that the intervention would be different for other LTCs. Additionally, the CYP determine their own intervention content.

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
Group Play Therapy	Nekah (2015)[132]	No	No manual. Includes four sessions of 'flexible play', which sounds like free play. Little detail provided.	Nothing suggested by data extraction form (check translation). Focus appears to be on MH, not LTC.
Group Play Therapy	Wang (2012)[137]	No	No manual. No suggestion of flexibility in delivery.	Nothing to suggest there is LTC-specific content/
Group Play Therapy	Zareapour (2009)[134]	No	Little detail, no manual.	Not enough detail, but sounds like generic play
Emotional intelligence training	Ashori (2013)[131]	No	No manual, structured intervention. No mention of how content was adapted for delivery to deaf children.	No LTC-specific content
Emotional intelligence training	Pourmoham adreza-Tajrishi (2013)[133]	No	No manual, structured intervention. No mention of how content was adapted for delivery to deaf children.	As in Ashouri (2013)

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
Relaxation	Bignall (2015)[118]	Yes	Script developed to train all CYP to practice exercises. Home-based practice.	Included asthma-specific guided imagery. Breathing exercises and PMR were not LTC-specific.
Relaxation	Yang (2004)[138]	No	All CYP listened to same audio tape to guide relaxation at home.	PRM only, no mention of asthma-specific content.
Stress management training	Boardway (1993)[119]	Yes	Structured intervention, but no manual. Individual diet plans introduced towards end of intervention.	Included content around the identification and management of diabetes-specific stressors. Included a three month programme focusing on treatment regimen adherence. Additional content focused on diet plans, insulin injection and timing related to meals etc.
Stress management training	Hains (2000)[121]	No	Structured intervention, but no manual. Al participants receive the same intervention.	Although activities such as identifying recent stressors would inevitably focus on LTC stressors, there is nothing to suggest activities could not be carried out in samples with other LTCs.
Music therapy	Bufalini (2009)[135]	No	All CYP go through the same process, but choose/make their own music.	Music therapy was arranged around a painful procedure for cancer. Could be applied for painful procedures related to other LTCs.

Intervention	Study	Content adapted to sample LTC?	Notes on delivery (e.g. was it manual-based, flexible delivery, individually-focused?)	Details of adapted components
Massage therapy	Diego (2001)[120]	No	Same intervention for all.	Although massage therapy was proposed to have physiological benefits for HIV (boosting immune function), the same massage could be delivered to CYP with other LTCs.
Heart Rate Variability Biofeedback	Yetwin (2011)[126]	No	Manualised protocol. No flexibility.	Nothing to suggest intervention couldn't be applied to other LTCs.
Resistance Training	Gordon (2010)[128]	No	All performed same exercises, but progressed in difficulty according to individual ability.	General exercise programme set to a level appropriate for the participants.

Appendix 6: Non-standardised meta-analyses

Non-standardised meta-analyses of outcomes following Parenting Programme with Acceptance and Commitment Therapy intervention

Brown et al. (2014) and Whittingham et al. (2014) used the same outcome measures to assess child behavioural problems, parenting styles and parent mental health, therefore non-standardised mean difference meta-analyses were performed on these outcomes (Figures Figure 25 to Figure 32). There was statistically significant improvements for the intensity (mean difference=27.5 points; 95% CI: 13.4 to 41.6 points, p<0.001) and number (mean difference=6.58 points; 95% CI: 1.50 to 11.7 points, p=0.01). Emotional difficulties improved by a mean of 1.21 points on the SDQ: Emotional symptoms subscale (95% CI: 0.38 to 2.05, p=0.004). There were also statistically significant improvements in parent mental health, as measured by the three DASS subscales: anxiety (mean difference=3.26 points; 95% CI: 0.71 to 5.81, p=0.01); depression (mean difference=3.75 points; 95% CI: 0.30 to 7.21, p=0.03); stress (mean difference=5.24 points; 95% CI: 2.13 to 8.35, p=0.001). There were similar statistically significant improvements in parenting laxness (mean difference=0.79 points; 95% CI: 0.36 to 1.21, p=<0.001) and over-reactivity (mean difference=0.77 points; 95% CI: 0.43 to 1.11, p=<0.001). There was less than 25% heterogeneity for all outcomes, with the exception of the ECBI: Problems scale, for which heterogeneity was moderate but not statistically significant (I^2=59.7%, p=0.12).

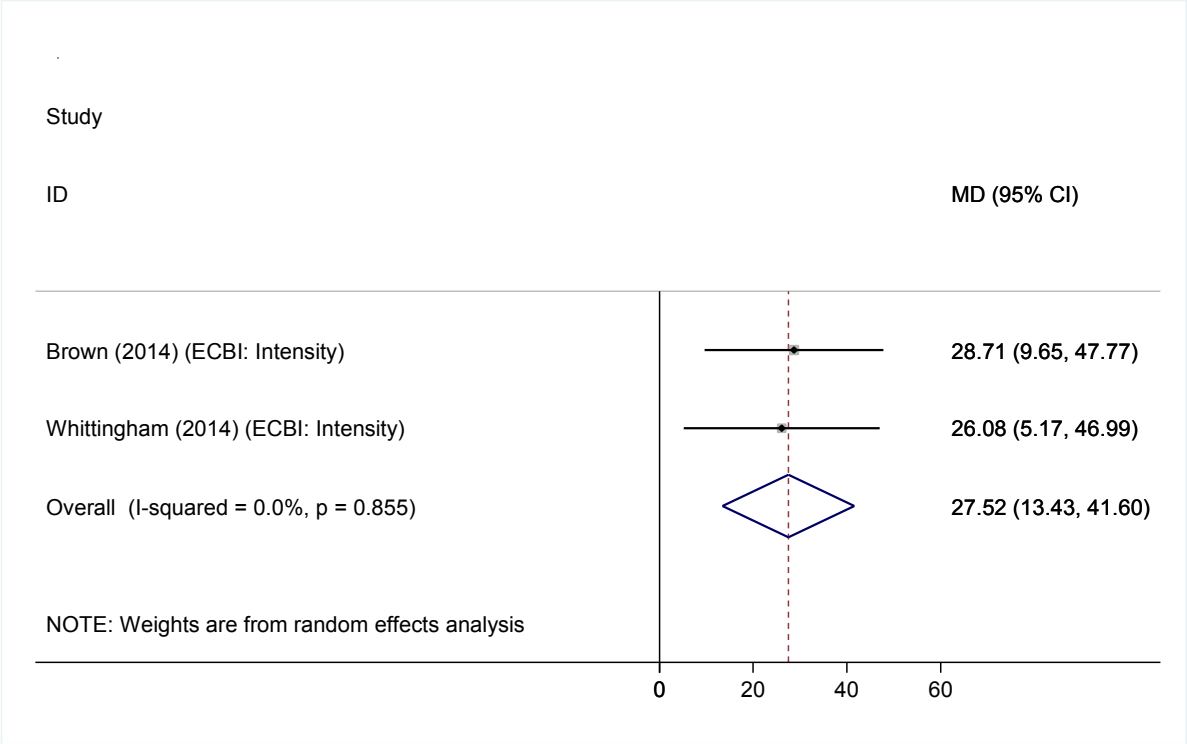

Figure 25. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on the intensity of child behavioural problems at post-intervention for included studies.

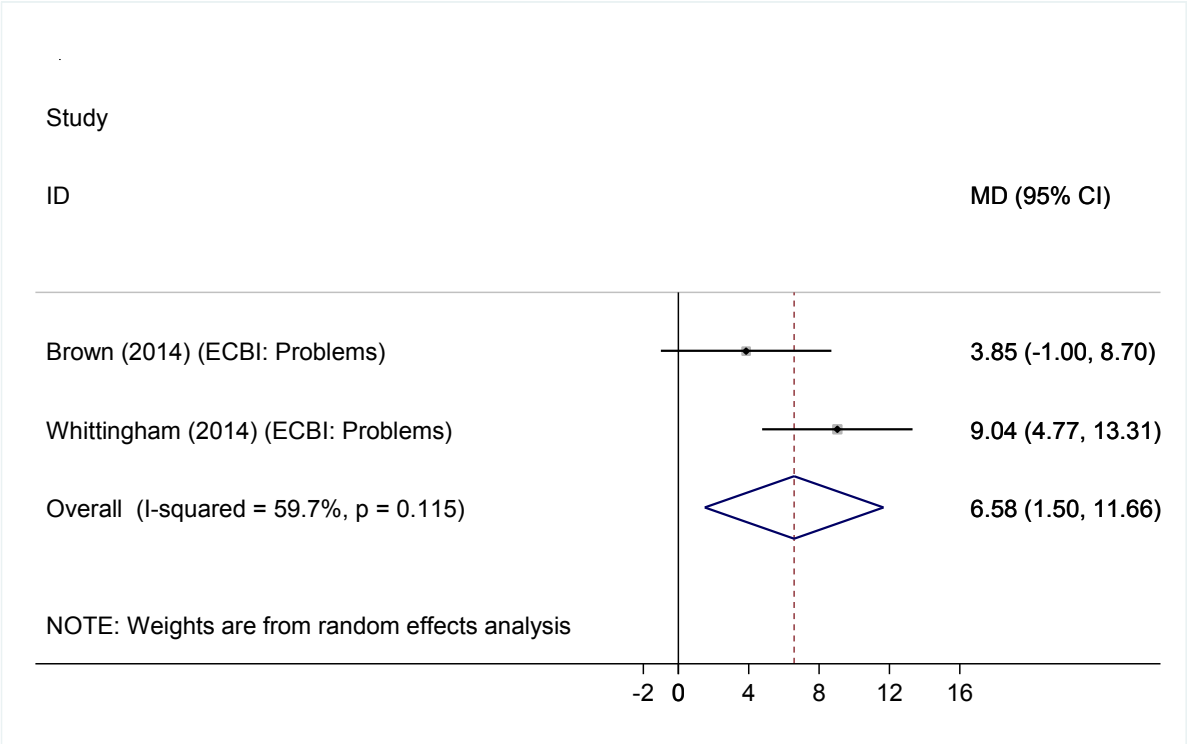

Figure 26. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on the number of child behavioural problems at post-intervention for included studies.

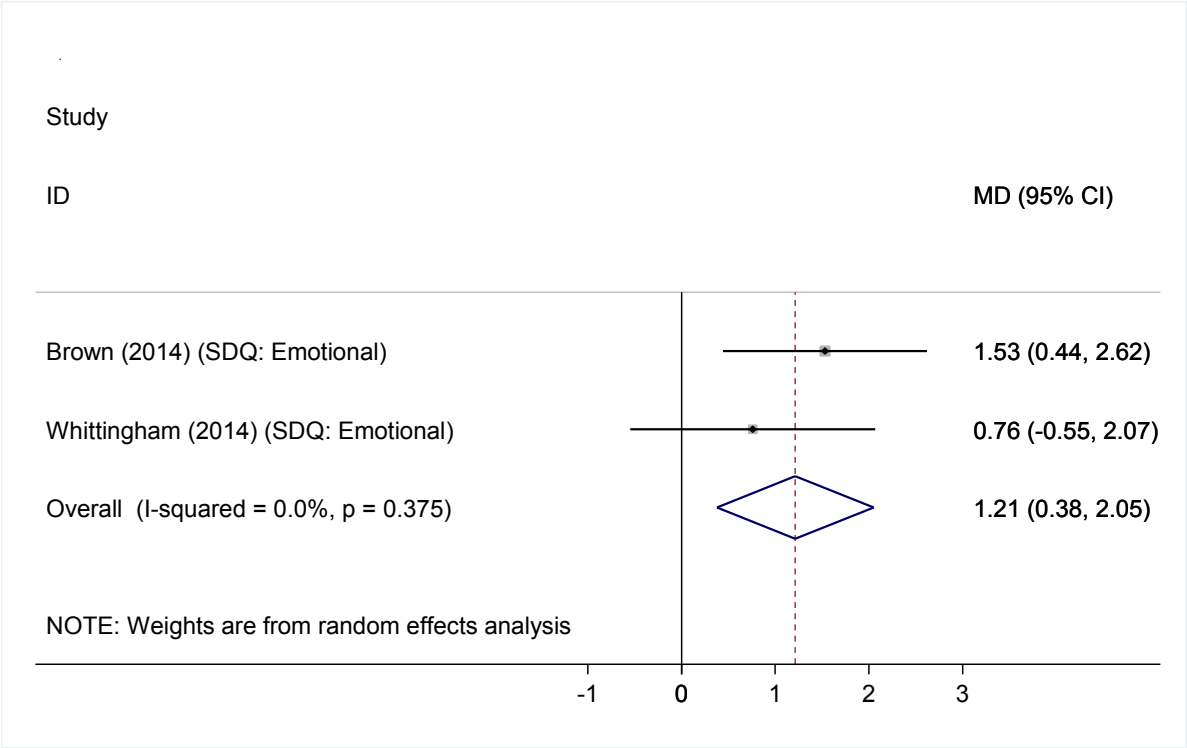

Figure 27. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on child emotional problems at post-intervention for included studies.

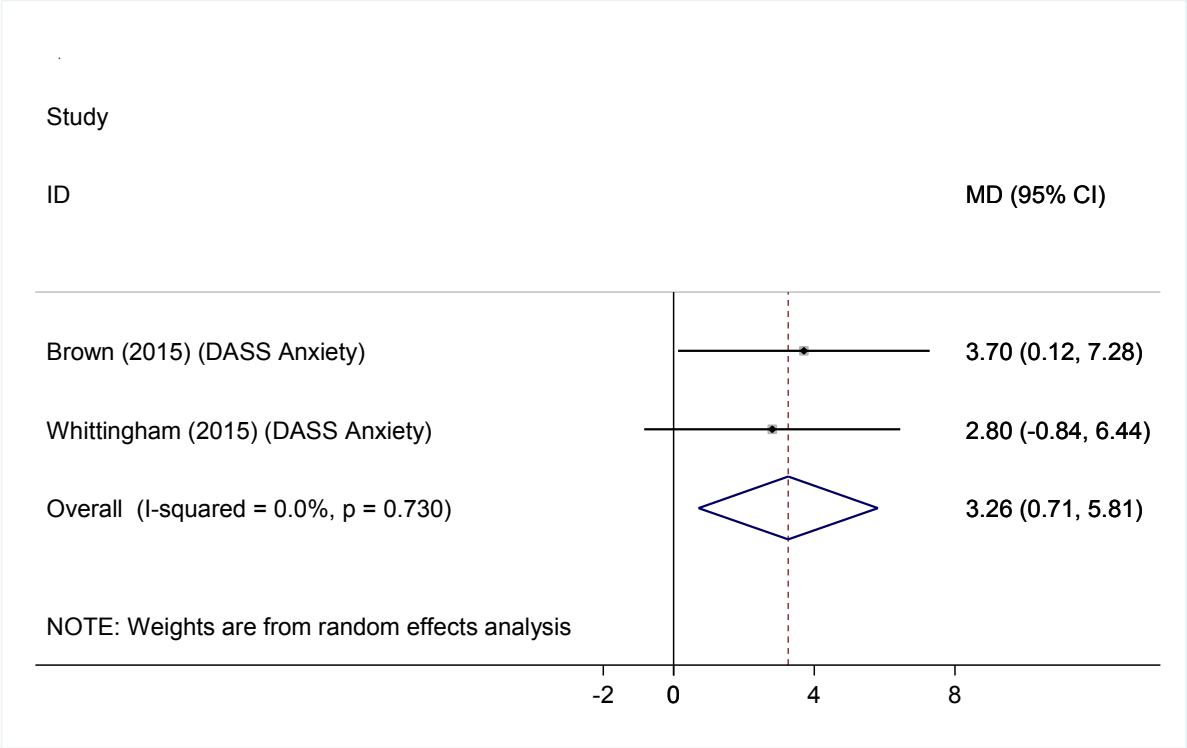

Figure 28. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parent anxiety at post-intervention for included studies.

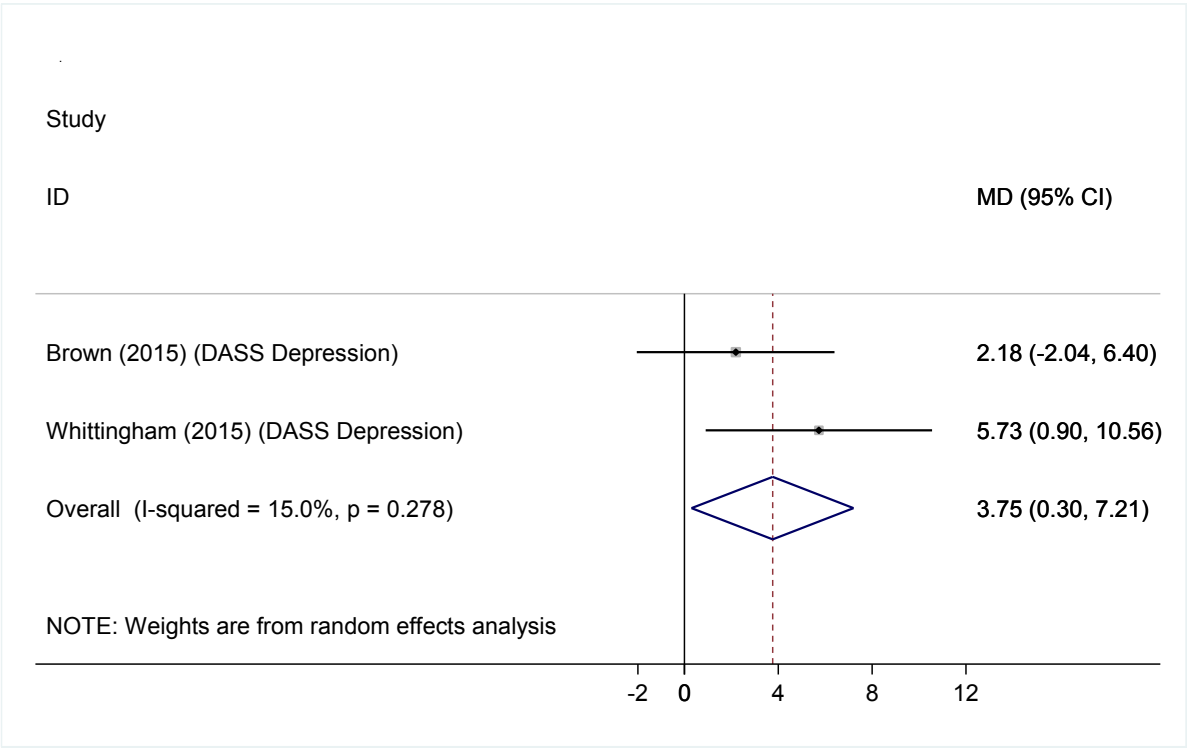

Figure 29. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parent depression at post-intervention for included studies.

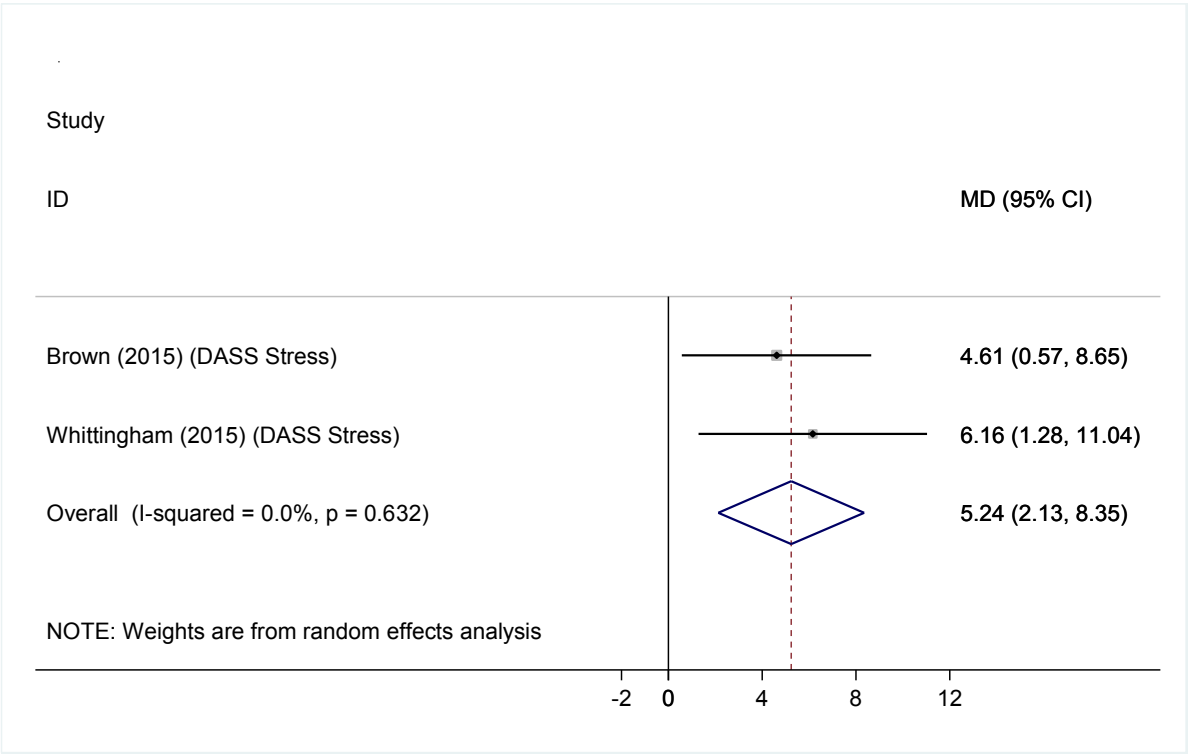

Figure 30. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parent stress at post-intervention for included studies.

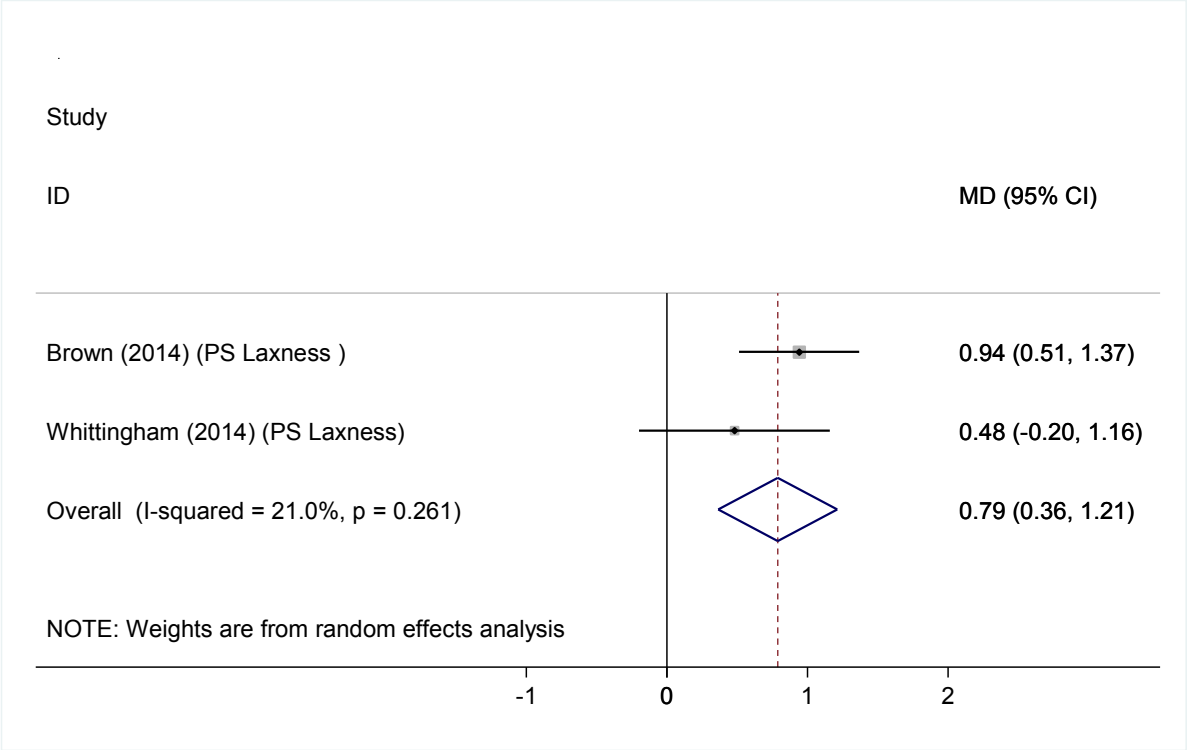

Figure 31. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parenting laxness at post-intervention for included studies.

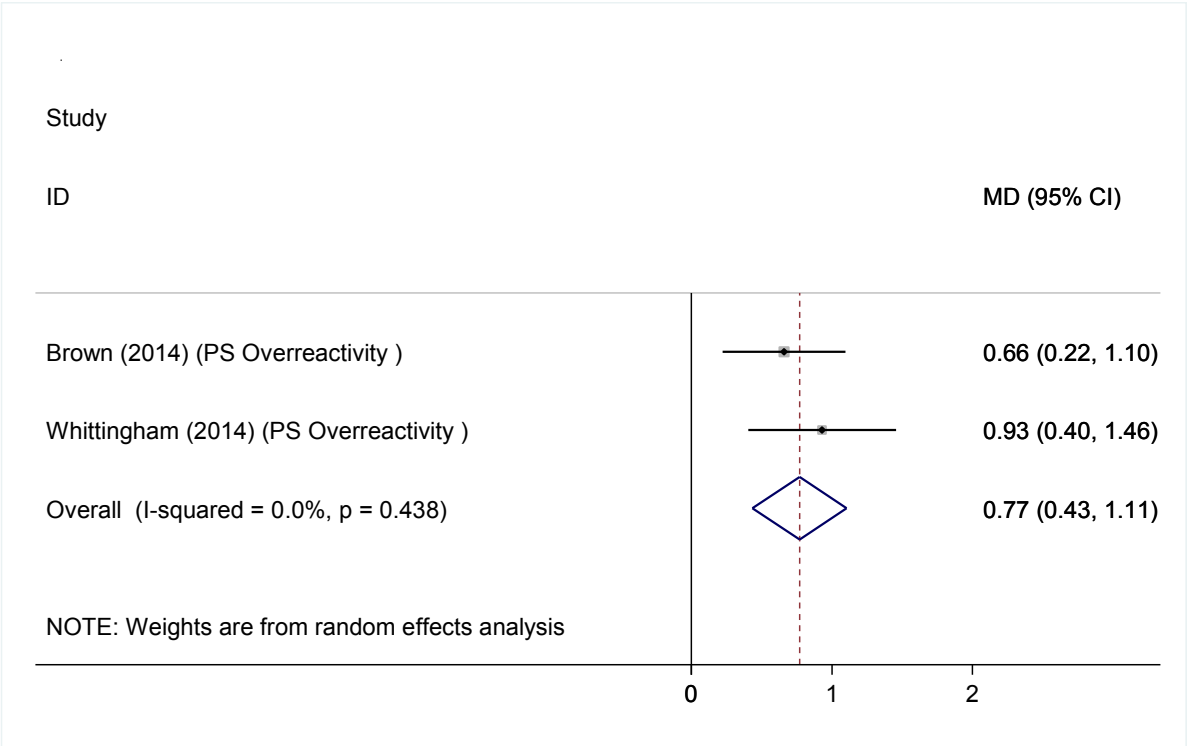

Figure 32. Forest plot showing the results of non-standardised meta-analysis of the effects of parenting programmes with acceptance and commitment therapy on parenting overreactivity at post-intervention for included studies.

Non-standardised meta-analyses of outcomes following Palliative Care intervention

Child physical functioning was assessed using the PedsQL: Physical functioning scale by both Lyon et al. (2014) and Shoshani et al. (2015), therefore a non-standardised mean difference meta-analysis was performed (Figure 33). Overall, there was a statistically significant 15.8 point increase on the scale following palliative care intervention (95% CI: 7.18 to 24.5, p<0.001).

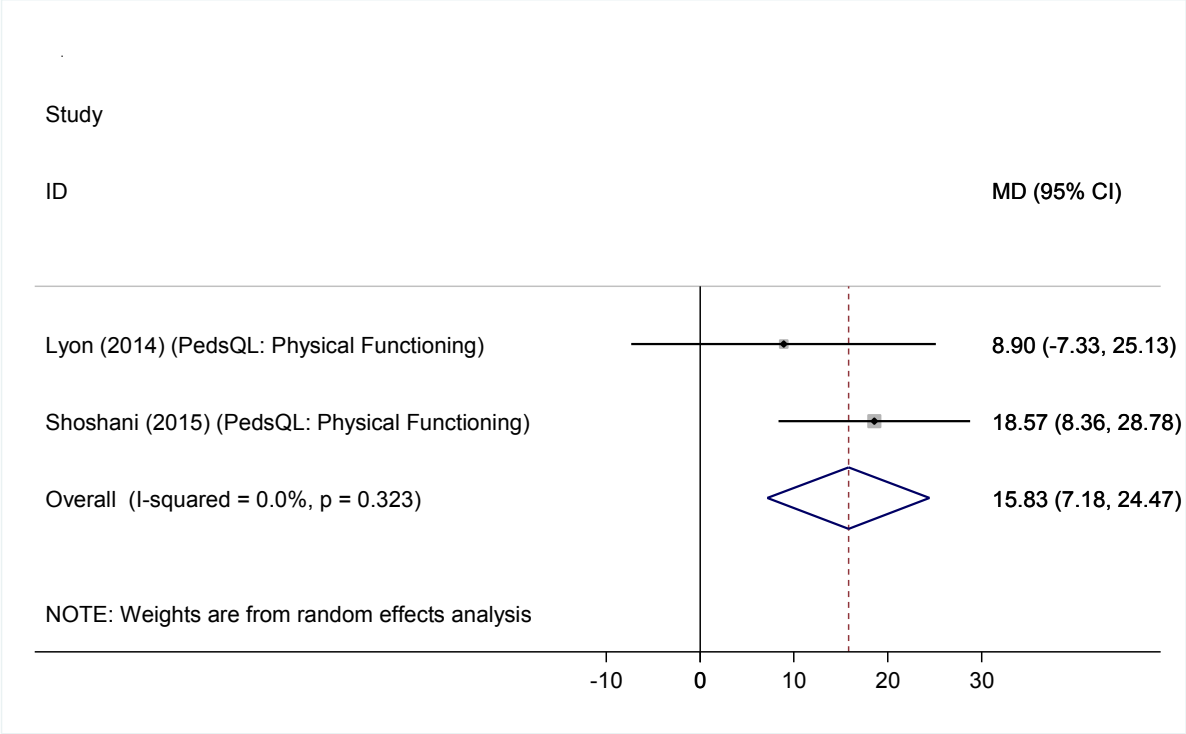

Figure 33. Forest plot showing the results of non-standardised meta-analysis of the effects of palliative care on child physical quality of life at post-intervention for included studies.

Non-standardised meta-analyses of outcomes following Stress Management Training intervention

LTC-specific stress was measured using the DSQ in the included studies that trialled stress management training (Board, Hains), therefore a non-standardised mean difference meta-analysis was performed on this outcome (Figure 34). There was an increase of 3.51 points on the measure, however this was not statistically significant, being associated with wide confidence intervals (mean difference=3.51; 95% CI: -18.4 to 25.4, p=0.75).

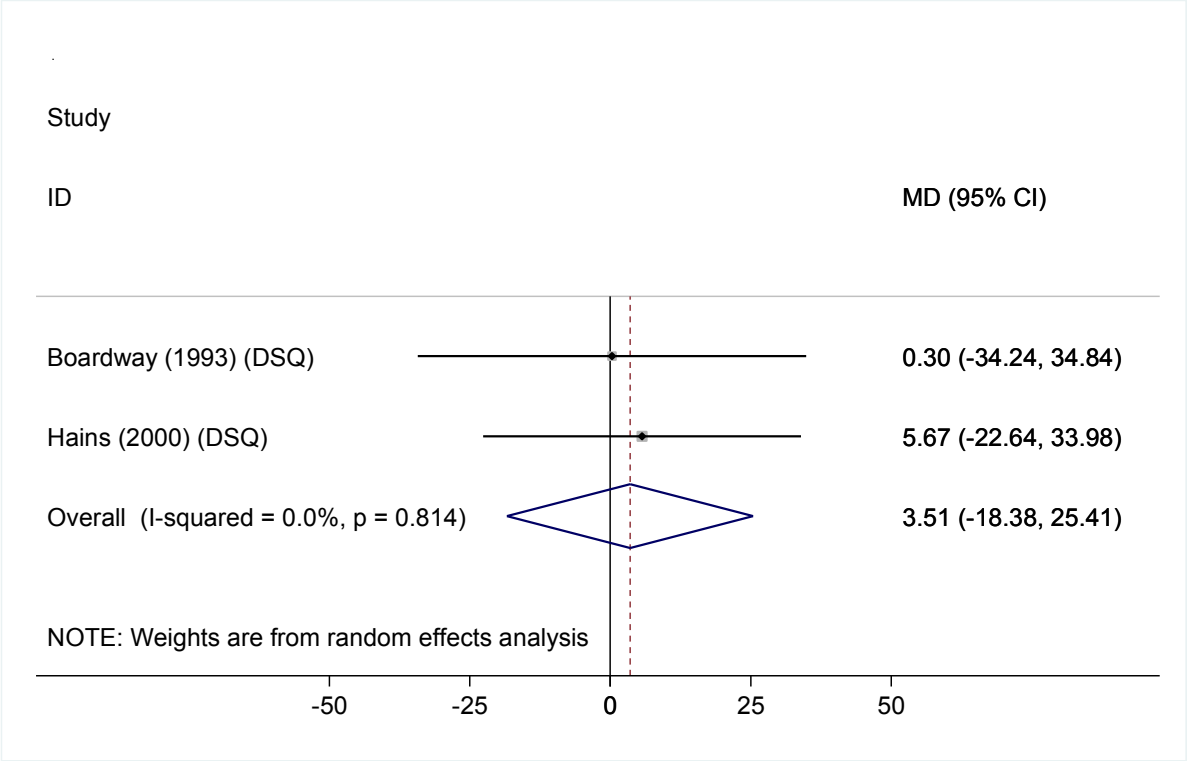

Figure 34. Forest plot showing the results of non-standardised meta-analysis of the effects of stress management training on child LTC-specific stress at post-intervention for included studies.

Appendix 7: Supplementary analysis – meta-analyses for cognitive behavioural therapy across long-term conditions

Where the same intervention, comparator and outcome category was used, meta-analysis was performed across LTCs. This opportunity was only presented for CBT across five LTCs.

Figure 1 shows a forest plot for the effect of CBT on outcomes at post-intervention across patients with epilepsy (Martinovic et al., 2006), chronic pain (Wicksell et al., 2009), persistent functional somatic complaints (PFSC, Masia Warner et al., 2011), inflammatory bowel disease (Szigethy et al., 2007) and diabetes (Serlachius et al., 2014). The plot is arranged by groups of outcomes, with depression, general mental health, LTC-specifc quality of life and LTC symptoms displayed. For depression, three measures (CES-D, BDI, HAMD) were combined to produce a single outcome for Martinovic et al. (2006). For LTC symptoms, parent and self-report of pain and the Children's Somatization Inventory were combined to produce a single outcome for Masia Warner et al. (2011), while five outcomes were combined to produce one single effect for Wicksell et al. (2009). These were the Pain and Impairment Rating Scale, pain intensity VAS, Pain Interference scale and Functional Disability Index (child and parent reports).

A statistically significant large beneficial effect was observed for depression (d=0.85; 95% CI: 0.50 to 1.19, p<0.001) and general mental health (d=0.83; 95% CI: 0.42 to 1.23, p<0.001) outcomes. In both cases, statistical heterogeneity was zero, providing further confidence in this effect. There was a large positive effect size for LTC-specific quality of life, however this was not supported at the 5% statistical significance level (d=0.88; 95% CI: -0.32 to 2.08, p=0.15). The wide confidence intervals and high heterogeneity (I^2=85.4%, p=0.009) reflect the imprecision and uncertainty for this outcome. LTC symptoms were improved with a medium sized statistically significant effect (d=0.61; 95% CI: 0.12 to 1.10, p=0.02), however the confidence intervals suggest uncertainty whether this effect is likely to be small or large. High heterogeneity further reflects the imprecision of this estimate (I^2=80.9%, p=0.005).

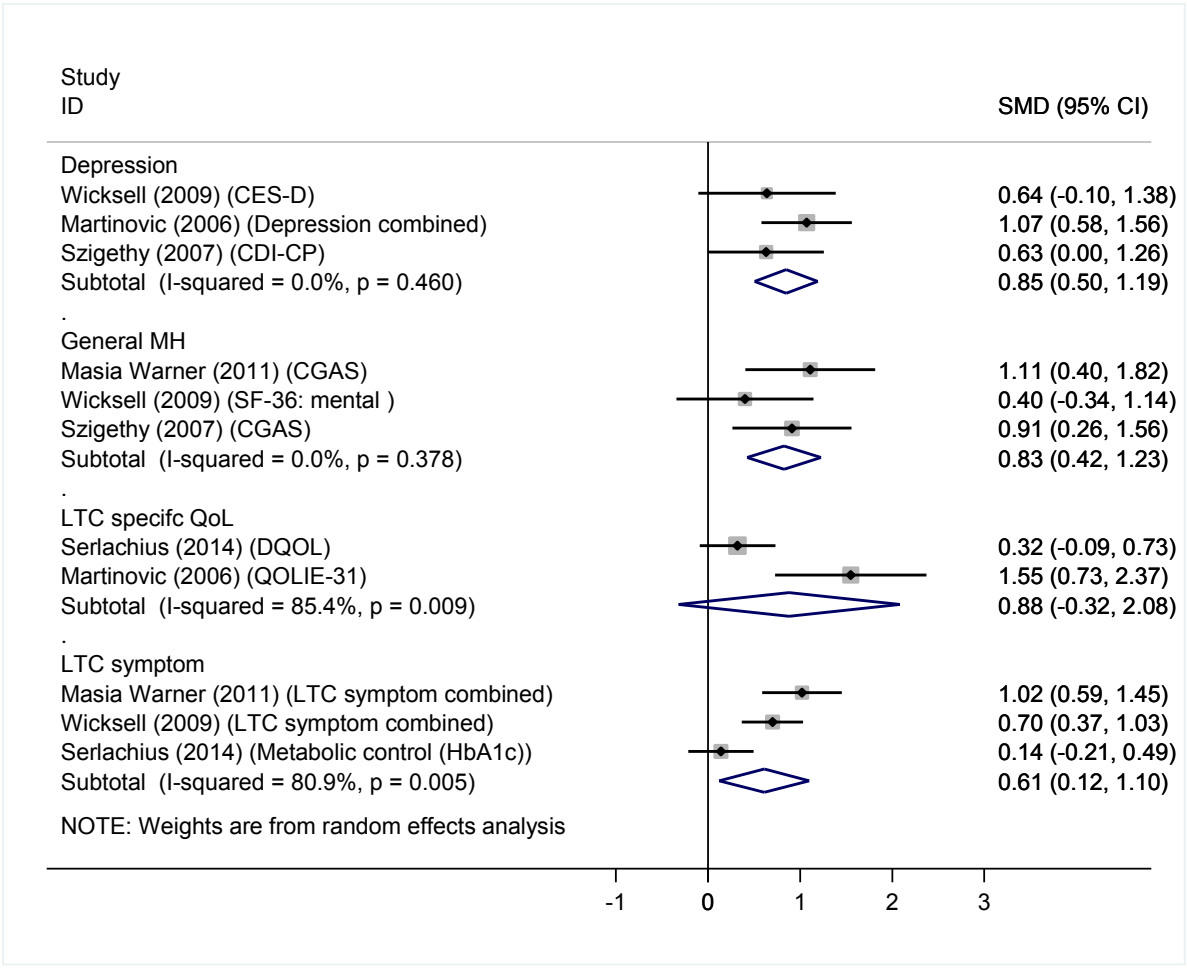

Figure 35. Forest plot showing the results of meta-analysis of the effects of CBT on depression, general mental health (MH), LTC-specific quality of life (QoL) and LTC symptoms at post-intervention for included studies.

SMD=Standardised Mean Difference (Cohen's d); CI=confidence interval; CES-D=Centre for Epidemiological Studies Depression scale; CDI-CP=Child Depression Inventory-Child and Parent version; CGAS=Children's Global Assessment Scale; DQOL=Diabetes Quality of Life scale; QOLIE-31=Quality of Life In Epilepsy Inventory-31 item version; LTC=Long Term Condition.

Appendix 8: Project team and expert advisory group who contributed to whole team meetings

Member	Area of expertise	Membership
Professor Rob Anderson	Health Economics	Project team
Dr Sophie Bennett		Project team
Professor Chris Dickens	Psychological Medicine	Project team
Professor Tamsin Ford	Child and adolescent mental health	Project team
Dr Ruth Garside	Evidence Synthesis	Project team
Professor Isobel Heyman	Consultant Child and Adolescent Psychiatrist	Project team
Paula Lavis	Coordinator Children and young people mental health coalition	Expert advisory group
Fiona Lockhart	Parent of a CYP with lived experience of an LTC and mental health difficulties	Expert advisory group
Professor Stuart Logan	Paediatric Epidemiology	Expert advisory group
Dr Darren Moore	Evidence synthesis	Project team
Dr Michael Nunns	Evidence synthesis	Project team
Morwenna Rogers	Information Specialist	Project team
Professor Roz Shafran	Clinical Psychology	Project team
Dr Liz Shaw	Evidence synthesis	Project team
Professor Jo Thompson Coon	Evidence synthesis	Project team

Member	Area of expertise	Membership
Dr Penny Titman	Consultant Clinical Psychologist	Project team
Professor Obioha Ukoumunne	Statistics	Project team
Professor Russell Viner	Adolescent health	Project team
Dr Erin Walker	Patient and Public Involvement Lead	Project team

Appendix 9: Search strategy used for Review 2 search with the PsycINFO (via OvidSp) database

#	Searches	Results
1	Chronic Disease/	0
2	Neoplasms/	28712
3	Diabetes Mellitus/	4470
4	Asthma/	4034
5	exp Respiratory Tract Diseases/	0
6	Cystic Fibrosis/	732
7	Cerebral Palsy/	4202
8	exp Epilepsy/	22725
9	exp Muscular Diseases/	0
10	exp Endocrine System Diseases/	0
11	exp Immune System Diseases/	0
12	exp HIV Infections/	0
13	exp Cardiovascular Diseases/	0
14	exp Nervous System Diseases/	0
15	exp Skin Diseases/	0
16	exp Digestive System Diseases/	0
17	exp Hematologic Diseases/	0
18	exp Otorhinolaryngologic Diseases/	0
19	exp Stomatognathic Diseases/	0
20	exp Eye Diseases/	0
21	exp Pain/	47653
22	Disabled Children/	0
23	((chronic* or longterm or long-term or "long standing" or physical) adj2 (condition* or ill* or disease* or disorder* or syndrome*)).ti,ab.	45152
24	(cancer* or neoplas* or tumor* or tumour* or malignan* or carcinoma* or "bone marrow" or leukaemia or leukemia).ti,ab.	63603
25	diabet*.ti,ab.	23493
26	asthma*.ti,ab.	6465
27	(respiratory adj2 (illness* or disease* or condition*)).ti,ab.	1201
28	cystic fibrosis.ti,ab.	993
29	cerebral palsy.ti,ab.	5460
30	quadriplegi*.ti,ab.	491

31	tetraplegi*.ti,ab.	291
32	diplegi*.ti,ab.	378
33	spinal cord injur*.ti,ab.	4595
34	muscular dystrophy.ti,ab.	1123
35	epilep*.ti,ab.	33297
36	seizure*.ti,ab.	25791
37	spina bifida.ti,ab.	868
38	(heart adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	9188
39	(cardiac adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	1390
40	(cardiovascular adj2 (condition* or disease* or disorder* or defect*)).ti,ab.	8891
41	(skin adj2 (condition* or disease* or disorder*)).ti,ab.	966
42	eczema.ti,ab.	330
43	(allergies or allergy).ti,ab.	1529
44	dermatitis.ti,ab.	532
45	(gastrointestinal adj (disorder* or disease*)).ti,ab.	669
46	((stomach or abdominal or gastrointestinal) adj pain).ti,ab.	1342
47	(bowel* adj2 inflammatory adj2 (condition* or disease* or illness*)).ti,ab.	602
48	(liver adj (disease* or transplant*)).ti,ab.	1621
49	hepatitis.ti,ab.	3515
50	(disabilit* adj5 child*).ti,ab.	13184
51	(human immunodeficiency virus or HIV).ti,ab.	42006
52	AIDS.ti,ab.	30925
53	(hyperthyroidism or hypothyroidism).ti,ab.	1635
54	an?emia.ti,ab.	1617
55	h?emophilia.ti,ab.	368
56	sickle.ti,ab.	1162
57	((renal or kidney) adj (disease* or disorder*)).ti,ab.	1900
58	nephrotic syndrome.ti,ab.	55
59	encephalomyelitis.ti,ab.	1604
60	chronic fatigue syndrome.ti,ab.	1946
61	((cleft or palate) adj lip).ti,ab.	295
62	craniofacial.ti,ab.	527
63	(deaf or deafness).ti,ab.	13257
64	(hearing adj (defect* or disorder*)).ti,ab.	347
65	blindness.ti,ab.	5420
66	((vision or visually or visual) adj (impaired or impairment*)).ti,ab.	4261

67	((persistent or chronic or recurring or frequent) adj (headache* or migraine*)).ti,ab.	1720
68	chronic pain.ti,ab.	11547
69	fibromyalgia.ti,ab.	2541
70	medically unexplained symptoms.ti,ab.	452
71	(spinal adj injur*).ti,ab.	301
72	or/1-71	335060
73	(child or children*).ti,ab.	529036
74	(adolescent or adolescents).ti,ab.	169875
75	teen*.ti,ab.	18103
76	(young adj (adult* or people)).ti,ab.	53922
77	youth*.ti,ab.	73029
78	73 or 74 or 75 or 76 or 77	701901
79	Mental Health/	47847
80	(psychological adj (illness* or disorder* or difficulties or problems or distress)).ti,ab.	23105
81	(mental adj (health or illness* or disorder* or distress or problem*)).ti,ab.	186832
82	(depression or depressed or depressive).ti,ab.	235411
83	((disruptive or challenging or antisocial) adj behavio?r).ti,ab.	11412
84	(behavio?r adj problem*).ti,ab.	12771
85	(anxiety or anxious).ti,ab.	157623
86	feelings.ti,ab.	57562
87	Internali*.ti,ab.	3
88	(wellbeing or well being).ti,ab.	61891
89	happiness.ti,ab.	11767
90	worry.ti,ab.	7030
91	distress.ti,ab.	48777
92	satisfaction.ti,ab.	82607
93	emotional.ti,ab.	174569
94	coping.ti,ab.	61721
95	or/79-94	828319
96	qualitative research/	6724
97	Ethnology/	1943
98	exp Questionnaires/	15996
99	phenomenology/	11126
100	Attitudes/	23102
101	interviewing/	3181
102	interview*.ti,ab.	250183

103	qualitative.ti,ab.	110757
104	(talked or asked).ti,ab.	81112
105	focus group*.ti,ab.	24246
106	ethnograph*.ti,ab.	20873
107	grounded theory.ti,ab.	11222
108	thematic.ti,ab.	14905
109	(barriers and (facilitators or enablers)).ti,ab.	2521
110	process evaluation.ti,ab.	1094
111	group discussion*.ti,ab.	7342
112	perception*.ti,ab.	232038
113	attitude*.ti,ab.	171590
114	views.ti,ab.	56281
115	experience*.ti,ab.	486970
116	or/96-115	1107429
117	intervention.ti,ab.	171181
118	psychotherapy.ti,ab.	77346
119	(support adj3 (group* or network)).ti,ab.	13125
120	therapy.ti,ab.	194016
121	counselling.ti,ab.	9299
122	peer support.ti,ab.	2673
123	social support.ti,ab.	35639
124	program*.ti,ab.	318905
125	(mental adj3 service*).ti,ab.	22848
126	training.ti,ab.	210131
127	technique*.ti,ab.	158051
128	treatment*.ti,ab.	508051
129	or/117-128	1226579
130	72 and 78 and 95 and 116 and 129	4011